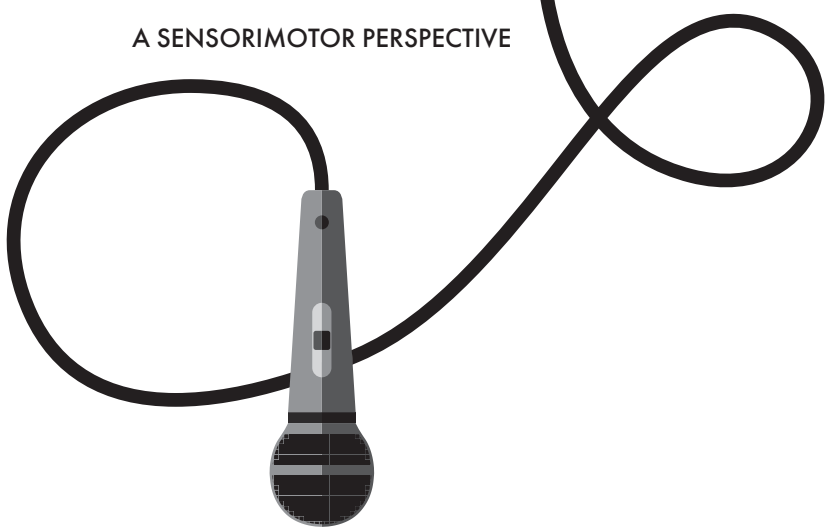
MARK R. VAN DEN BUNT

Revisiting the phonological deficit hypothesis in dyslexia:

A SENSORIMOTOR PERSPECTIVE



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A sensorimotor perspective

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Mark Ruben van den Bunt
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Promotor:
Prof. dr. L.T.W. Verhoeven

Copromotor:
Dr. M.A. Groen

Manuscriptcommissie:
Prof. dr. J.P.M. Fikkert
Prof. dr. A.H. Neijt
Prof. dr. P. Ghesquière (KU Leuven, België)

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Mark Ruben van den Bunt
Born on March 27, 1989
in Amersfoort (The Netherlands)

- -

Supervisor:
Prof. dr. L.T.W. Verhoeven

Co-supervisor:
Dr. M.A. Groen

Doctoral Thesis Committee:
Prof. dr. J.P.M. Fikkert
Prof. dr. A.H. Neijt
Prof. dr. P. Ghesquière (KU Leuven, Belgium)

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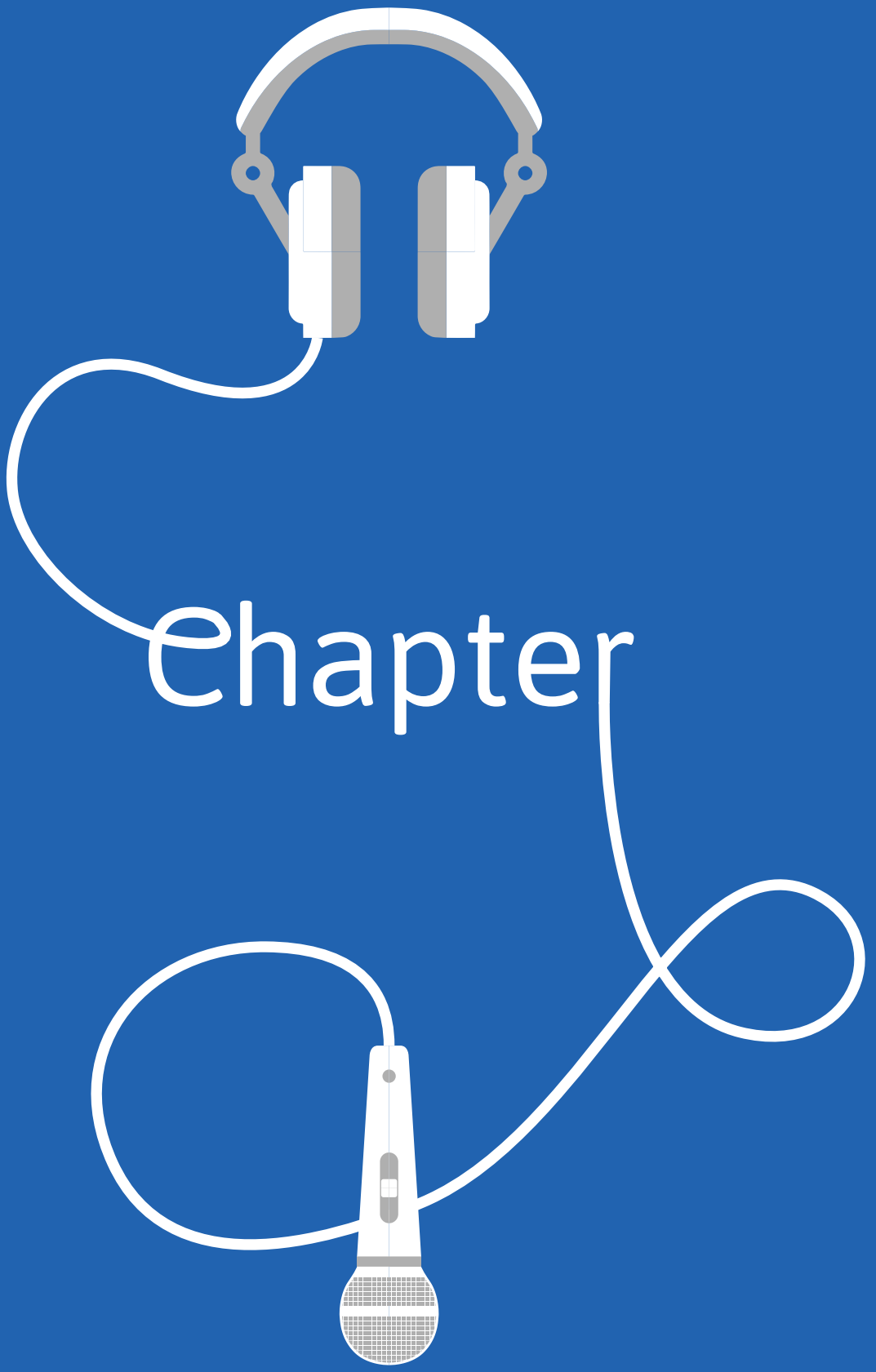
Chapter

GENERAL INTRODUCTION

In today's society, reading is one of the most fundamental cognitive skills, and hence is a strong predictor of academic and vocational success (Bashir & Scavuzzo, 1992; Finucci, Gottfredson, & Childs, 1985). Proficient readers are usually able to identify words within 200 milliseconds and do so without much effort. What often is not realized is that reading is a complex ability that, during development, is built on (speech) motor and linguistic skills. Importantly, humans are not genetically hardwired for reading: reading is a fairly recent cultural invention. Unlike learning to speak, which occurs automatically, learning to read has been called an *'unnatural act',* which requires explicit instruction (Gough & Hillinger, 1980; Liberman, 1989). Adequate speech skills are essential in the process of learning to read, and speech difficulties have been associated with reading problems (Lyytinen et al., 2004; Lyytinen, Poikkeus, Laakso, Eklund, & Lyytinen, 2001; Scarborough, 1990).

A dominant hypothesis in reading research is that the quality of phonological representations is related to reading skill and development and poorly defined representations are recurrently hypothesized to be a causal marker of dyslexia (Liberman, Shankweiler, & Liberman, 1989; Elbro, 1998; Boada & Pennington, 2006). However, the precise nature and origin of phonological deficiencies in dyslexia is hotly debated. In the current dissertation, I propose that, in order to make progress in understanding how phonological representations are related to reading, sensorimotor control of speech should be examined. In the speech production literature, sensorimotor control of speech is frequently hypothesized to be crucially involved in de development and maintenance of adequate phonological representations (Guenther, Ghosh, & Tourville, 2006; Tourville & Guenther, 2011). The aims of the present dissertation are to examine (i) whether dyslexia is characterized by impaired sensorimotor control; (ii) whether neurobiological measures support the importance of speech sensorimotor control in reading skill; (iii) whether sensorimotor control of speech explains individual differences in reading skill in individuals with dyslexia and (iv) how the sensorimotor control of speech relates to early reading development in typically developing children. In this general introduction, I will first describe the cognitive foundations of reading and provide an overview the most relevant theories of dyslexia. Secondly, the neurobiological basis of reading and dyslexia will be outlined. Thirdly, I will describe the rationale of the present dissertation, including why measures of the sensorimotor control of speech are crucial to make progress in understanding differences in reading skill. Lastly, I will provide an outline of the Chapters of this dissertation.

COGNITIVE FOUNDATIONS OF READING AND DEVELOPMENTAL DYSLEXIA

Children typically speak and combine words prior to the age of two years (Zubrick, Taylor, & Rice, 2007), but the understanding of the phonological structure of the language continues to develop during childhood. More specifically, children start with whole-word representations of language at age one to two (Ferguson & Farwell, 1975) and increasingly segmentalized phonological representations develop gradually as the child's lexicon grows until the age of eight years (Fowler, 1991). In an alphabetic language, the written symbols refer to these smaller segments of speech and a child's phonological development is an important building block for reading development. Learning to read is about mastering the alphabetic principle, the rules that link phonological to orthographic representations. In the early stages of reading, word reading predominantly occurs via a system of grapheme-phoneme conversions that allows written information to be spoken. This 'phonological-recoding' strategy is necessary to decode orthographically unfamiliar words. In contrast, fluent and automatic reading of text is hypothesized to largely take place via a direct orthographic-to-meaning route (Plaut, McClelland, Seidenberg, & Patterson, 1996; Seidenberg & McClelland, 1989). A subset of the population experiences difficulties in establishing adequate phonological recoding skills and/or orthographic-to-meaning links, and are diagnosed with developmental dyslexia (dyslexia hereafter). Dyslexia is the most common neurodevelopmental disorder—with a prevalence estimate of approximately seven percent across languages (Goswami, 2015)—characterized by persistent difficulties in fluent and/or accurate word reading, despite adequate intelligence and reading instruction (Lyon, Shaywitz, & Shaywitz, 2003). It should be noted that research suggests that this disorder represents the lower tail of normally distributed reading skills and that any cutoff for diagnosing dyslexia remains arbitrary. Nevertheless, reading has become increasingly important for academic and vocational success (Bashir & Scavuzzo, 1992; Finucci et al., 1985) and economic costs of literacy problems for society are significant. For instance, recent calculations estimate costs of low literacy skills to approach one billion per annum in the Netherlands (PriceWaterhouseCoopers, 2013). Despite decades of research into the neurobiological and cognitive origins of dyslexia, the underlying cause(s) is still hotly debated. There is large consensus that dyslexia is characterized by deficiencies in phonological processing. However, several competing theories have been proposed to explain the underlying origin of these phonological deficiencies. The studies presented in the current dissertation have implications for several theories on the origins of dyslexia, which will be discussed below. Importantly, these theories can be divided in two 'categories'. First, the phonological deficit theory,

the allophonic mode of speech perception theory, and the phonological access theory all pose that dyslexia is primarily characterized by difficulties in speech processing. The other theories (referring to general auditory processing, cerebellar, statistical learning and magnocellular deficits) do not reject speech impairments in dyslexia, but claim that these deficits are secondary to broader impairments. Theories that will not be addressed by the experiments of the presented studies, such as exclusively visual theories (e.g. binocular instability; Jainta & Kapoula, 2011), will be omitted from the following overview.

Speech-specific theories on dyslexia

One of the most dominant theories of dyslexia is the phonological deficit theory (Boada & Pennington, 2006; Høien-Tengesdal & Tønnessen, 2011; Snowling, 1981). This theory claims that deficient phonological representations underlie the reading deficit in individuals with dyslexia. The reasoning is usually as follows: learning to read requires the acquisition of grapheme-phoneme correspondences (Liberman, Shankweiler, & Liberman, 1989); if phonological representations are fuzzy, it hampers the ability to establish stable and automatized phoneme-grapheme correspondences (Elbro, 1998). This theory is largely based on the consistently poorer performance of individuals with dyslexia on measures of phonological awareness—the ability to access and manipulate individual sounds in words (Melby-Lervag, Lyster, & Hulme, 2012)—and, more controversial, on measures of speech perception (Hakvoort et al., 2016; Ziegler, Pech-Georgel, George, & Lorenzi, 2009) and production (Catts, 1997; Smith, Roberts, Lambrecht-Smith, Locke, & Bennett, 2006). Impaired phonological awareness has been reported in children (e.g. Carroll & Snowling, 2004) and in adults (e.g. Nergård-Nilssen & Hulme, 2014) with dyslexia. In addition, training phonological awareness skills prior to formal literacy instruction is associated with better subsequent reading skills (Bus & van IJzendoorn, 1999), which suggests a causal role of phonological awareness in literacy development. However, tasks measuring phonological awareness rely on many cognitive abilities, including attention and working memory, and are hence not specific enough to claim that impaired phonological representations underlie dyslexia. In contrast, speech perception and speech production directly reflect the quality of phonological representations, but the evidence for speech perception and/or production deficits in individuals with dyslexia is equivocal. For instance, speech perception is reported to be deficient in dyslexia, however, this might not be the case for all phonetic contrasts (Cornelissen, Hansen, Bradley, & Stein, 1996) and not in all conditions (i.e. in silent vs. in noise conditions (Hazan, Messaoud-Galusi, & Rosen, 2012). Considering that phonological representations are not merely perceptual, they also include motor commands associated with the representation, the scant attention

for speech production in dyslexia is striking. The studies that do measure the quality of speech production in individuals with and without dyslexia suggest that dyslexia is characterized by impaired oral motor skills (Malek, Amiri, Hekmati, Pirzadeh, & Gholizadeh, 2013) and articulatory skills (Catts, 1997).

An alternative explanation for the weaker perceptual discrimination between phonemes in individuals with dyslexia is that dyslexia is characterized by a higher sensitivity for phonetic distinctions that are not relevant in the native language, labeled an allophonic mode of speech perception (Serniclaes, Van Heghe, Mousty, Carré, & Sprenger-Charolles, 2004). According to this theory, an allophonic mode of perception obstructs the development of robust phonemic representations that can be considered a prerequisite for setting up adequate phoneme-grapheme correspondences. A core idea that founded this hypothesis is that newborns appear to be able to discriminate between all/most existing phonetic contrasts and that with development, infants lose the ability to discriminate between sounds that are not present in the native language (Aslin, Pisoni, Hennessy, & Perey, 1981; Werker & Tees, 1984). Individuals with dyslexia possibly stay (longer) in this initial, universal stage of phonology in which the speech perception system is not (yet) adequately attuned to the phonetic contrasts present in the native language. The findings that many children at risk of dyslexia (Noordenbos, Segers, Serniclaes, Mitterer, & Verhoeven, 2012), children diagnosed with dyslexia (Serniclaes et al., 2004) and adults diagnosed with dyslexia (Noordenbos, Segers, Serniclaes, & Verhoeven, 2013) demonstrate an allophonic mode of perception, suggest that this allophonic mode of perception might be a persistent condition in dyslexia. One of the predictions from this theory is that individuals with dyslexia are better at discriminating between foreign speech sounds with boundaries that are not present in the native language (Serniclaes et al., 2004). However, a study that explicitly examined this prediction in French adults did not find any differences in the perception and production of non-native speech sounds between adults with and without dyslexia (Soroli, Szenkovits, & Ramus, 2010). Relatedly, individuals with dyslexia appear to be worse in categorizing talker dialects (Long, Fox, & Jacewicz, 2016), which further questions a universal mode of speech perception.

A recently popular view on the reported phonological deficiencies in dyslexia comes from Ramus and Szenkovits (2008). In a reinterpretation of an abundance of data on the phonological deficit, they propose that the deficit could best be described as a deficit in access to phonological representations, rather than a deficit in the representations themselves. This 'phonological access' theory suggests that the phonological deficit becomes particularly apparent when tasks require conscious awareness of the

phonological structure of speech and place a strong demand on short-term memory and fluent/automatized access to phonological representations (Boets et al., 2013; Long et al., 2016; Ramus & Szenkovits, 2008)

General theories on dyslexia

Alternative, non-speech specific theories of dyslexia, do not question impaired functioning of individuals with dyslexia on tasks that measure phonological abilities, they rather propose that this phonological deficit is secondary to more general sensory or cognitive impairments (Goswami, 2015; Ramus et al., 2003). Several 'variants' of this idea have been proposed throughout the literature. For instance, the rapid auditory processing theory (Marshall, Snowling, & Bailey, 2001; Tallal, 1980), proposed that children with reading impairments show deficiencies in the rate at which they can process auditory information. Rapid processing of auditory stimuli was argued to be associated with reading as it is was hypothesized to be an important skill for phoneme perception (Tallal, 1980). A distinct, but related, hypothesis states that dyslexia might be characterized by impaired detection of the amplitude and frequency modulation of speech signals that hamper the adequate segregation of syllable onsets (Goswami et al., 2002). The awareness of the syllabic structure of the language precedes the awareness of phonemes and a deficit in segregating syllables could delay and affect the development of the entire phonological system (Goswami et al., 2002). The evidence for these theories is also mixed. First, studies that do report deficits in general auditory perception in individuals with dyslexia, also indicate that the majority of participants perform within the range of typical readers (Ramus et al., 2003; Tallal, 1980). Moreover, some studies are not able to replicate deficiencies in auditory processing and point to strategic and cognitive abilities that may affect performance on these low-level auditory measures (Marshall et al., 2001; Ramus et al., 2003). Furthermore, although measures of lower-level auditory skills have been reported to differ between groups of typical and dyslexic readers, these measures do not account for much variance in reading skill (Heiervang, Stevenson, & Hugdahl, 2002). Finally, auditory deficits do not consistently predict deficits in phonological representations (Mody, Studdert-Kennedy, & Brady, 1997; Share, Jorm, Maclean, & Matthews, 2002).

Another domain-general theory that attempts to explain the full range of presented deficits in dyslexia, is the cerebellar deficit hypothesis (Nicolson, Fawcett, & Dean, 2001). This theory claims that dyslexia can be characterized by difficulties in skill automatization and motor control. Individuals with dyslexia are reported to show impairments in balance and muscle tone (Fawcett & Nicolson, 1999), and decreased functional activation in cerebellar areas when performing a motor task, hypothesized to be cerebellar-driven

(Nicolson et al., 1999, 2001; Rae et al., 1998). Motor control, obviously, is involved in speech articulation and this theory suggest that impairments in speech articulation lead to deficient phonological representations. Additionally, the cerebellum is also assumed to be critically involved in the automatization of motor behaviors. Reading, and more specifically converting graphemes in phonemes, is strongly dependent on these automated behaviors (Nicolson et al., 2001; Ramus et al., 2003).

The magnocellular theory of dyslexia finds its origin both in early neuroanatomical work and in research in the visual domain that showed a reduced activation in visual areas in individuals with dyslexia in response to visual motion perception (Eden et al., 1996). This theory derives its name from the cells that are relatively large (i.e. *magno*) and sensitive to light and fast motion in a relatively large area of the retina (Kaplan & Shapley, 1986). Early neuroanatomical work in individuals with dyslexia showed disordered magnocellular layers in the lateral geniculate nucleus (Galaburda & Livingstone, 1993), an important relay system for the processing of visual information. With respect to reading, an impaired visual magnocellular may affect the ability to direct visual attention and eye movements involved in reading (Stein, 2001). Although rooted in research on the visual system in dyslexia, the magnocellular theory has been extended to be domain-general and poses that a general sensory deficit, driven by magnocellular systems, might underlie dyslexia (Stein, 2001). Moreover, since the cerebellum relies, amongst others, on input from these magnocellular systems, a dysfunctional cerebellum does fit into this hypothesis (Ramus, 2003). In short, this magnocellular theory unifies the general auditory, cerebellar, and visual theories of dyslexia and points to a single underlying mechanism. This unification of several theories, however, comes at a price: it also imports the reported shortcomings of general auditory and cerebellar theories. For instance, findings are not consistently replicated or do not apply to the majority of the individuals with dyslexia. Moreover, early data on a magnocellular deficit in dyslexia relied on designs with individuals with dyslexia and chronological age matched controls (Livingstone, Rosen, Drislane, & Galaburda, 1991; Lovegrove, Bowling, Badcock, & Blackwood, 1980). Since reading experience differs strongly between these groups, it is possible that magnocellular differences are a consequence, rather than a cause of dyslexia (Goswami, 2015).

Lastly, it has been proposed that dyslexia is not (primarily) characterized by sensory deficits, but that deficient statistical/implicit learning underlies the disorder (Vicari et al., 2005). Statistical learning is thought to be crucially involved in language learning by means of extracting statistical regularities from speech/print and use this knowledge to learn, for instance phonotactic patterns (Dell, Reed, Adams, & Meyer, 2000) and phonemic categories (Maye, Weiss, & Aslin, 2008). With respect to dyslexia, individuals

with dyslexia could be worse in extracting statistical knowledge of letter patterns (Rothe, Schulte-Körne, & Ise, 2014) and/or learning grapheme-phoneme correspondences (Apfelbaum, Hazeltine, & McMurray, 2013). Statistical learning is often hypothesized to be domain-general (Kirkham, Slemmer, & Johnson, 2002; Pacton, Fayol, & Perruchet, 2005), although some studies also describe modality specific effects in statistical learning (Conway & Christiansen, 2005; Siegelman & Frost, 2015). Several studies do report group differences between individuals with and without dyslexia for statistical learning (e.g. Pavlidou, Louise Kelly, & Williams, 2010; Stoodley, Harrison, & Stein, 2006), however, a similar number of studies failed to find differences (Laasonen et al., 2014; Nigro, Jiménez-Fernández, Simpson, & Defior, 2016). A recent meta-analysis suggests that there is no evidence for a strong claim of impaired statistical learning in dyslexia (Schmalz, Altoè, & Mulatti, 2016).

In short, several competing theories on the underlying causes of dyslexia exist. These theories can be roughly divided in speech-specific and general theories. Speech specific theories claim that the main cause of dyslexia is rooted in deficient (access to) phonological representations, domain-general theories claim that deficits in phonological representations are secondary to general sensory or cognitive deficits. The studies in the present dissertation address, amongst others, the criticism on phonological representation theories that deficits in phonological representations become particularly apparent when tasks demand meta-phonological knowledge, working memory and conscious access, while the representations themselves might be intact. The implications of the studies described in this dissertation will be further discussed in the final Chapter 6.

NEUROBIOLOGICAL FOUNDATIONS OF READING AND DYSLEXIA

In addition to studies that examined behavioral correlates of reading development and dyslexia, many studies have been devoted to understanding the neurobiological basis of reading and dyslexia. Neuroimaging has further corroborated the existence of two complementary (phonological recoding and orthographic-to-meaning) routes of reading and extended our understanding of what goes awry in dyslexia. Functional neuroimaging studies have demonstrated that reading is largely lateralized to the left hemisphere (Price, 2012), and that three important foci compose a distributed network for reading: the fusiform gyrus, temporo-parietal systems and inferior frontal systems (Jobard, Crivello, & Tzourio-Mazoyer, 2003; Pugh et al., 2000, 2013). The left fusiform gyrus is systematically activated for print stimuli, independent of writing system (Wong, Jobard, James, James, & Gauthier, 2009), and has been labeled the *visual word form area*

(Cohen et al., 2000; Dehaene & Cohen, 2011). Orthographic information, processed in the visual word form area, is hypothesized to be mapped onto auditory (temporo-parietal) and semantic (frontal) representations. The temporo-parietal systems are usually associated with phonological processing and the conversion from orthography to phonology (Shaywitz & Shaywitz, 2008). The left inferior frontal system comprises Broca's and premotor areas (Shaywitz & Shaywitz, 2008) and this system is hypothesized to be involved in speech and print articulation. A recent account on how reading is parasitic on the neural circuitry for speech comes from Rueckl and colleagues (2015). In that study, it was shown, that reading skill across four different orthographies was particularly explained by how the neural circuitry for reading and the circuitry for speech converged, suggesting that the reading network is building on, and constrained by, the brain organization for speech.

In terms of the development of the neural circuitry for reading, several studies suggest that learning to read is associated with a disengagement of the right hemisphere for orthographic stimuli and developing a highly-specialized left-hemispheric neurocircuitry for reading (Orton, 1937 in Turkeltaub, Gareau, Flowers, Zeffiro, & Eden, 2003), with increasing activation during reading for the left fusiform gyrus and temporo-parietal systems as a function of reading skill (Shaywitz et al., 2002; Turkeltaub et al., 2003). A more intrinsic developmental account on reading development comes from Pugh and colleagues (Pugh et al., 2000, 2010), posing that early in development, left and right hemisphere temporal and parietal areas operate in conjunction with frontal areas to do the computational work to develop phonological awareness and to subsequently encode relations between orthographic, phonological and semantic representations that ultimately result in a left specialized circuitry that supports fluent word recognition. Based on this account, bilateral activations during word reading are expected for beginning readers, but as readers develop, this activation is expected to become less distributed. Another perspective on reading development poses that becoming a mature reader is associated with a shift from using a dorsal route of reading towards a ventral route (Pugh et al., 2000; Shaywitz et al., 2002). The dorsal route is thought to underlie explicit and effortful grapheme-to-phoneme conversions whereas the ventral route supports automatized whole word recognition.

With respect to structural brain basis for reading, many studies have used diffusion tensor imaging (DTI) to examine whether the integrity of white matter tracts are associated with reading skill. These DTI studies repeatedly showed white-matter integrity with reading associations in tracts that connect key regions that compose the reading network (Beaulieu et al., 2005; Klingberg et al., 2000; Niogi & McCandliss, 2006). Tracts of particular interest in this respect are the inferior fronto-occipital fasciculus

and the arcuate fasiculus. The former tract connects visual (occipital) areas with speech articulatory (inferior frontal) areas and is associated with more advanced reading in which words are instantly recognized via orthographic representations (Pugh et al., 2001; Vandermosten et al., 2015). The arcuate fasciculus connects auditory (temporo-parietal) areas with speech articulatory (inferior frontal) areas and this tract is frequently hypothesized to be associated with the beginning stages of reading development and to underlie the process of converting graphemes to phonemes (Pugh et al., 2010). Although this neurodevelopmental model is influential in the literature, it is not clearly backed up by empirical data. Recent studies suggest that both the left inferior fronto-occipital and the left arcuate fasciculus, as well as their right hemisphere homologues are related to early reading development (e.g. Vandermosten et al., 2015).

With respect to dyslexia, the three regions that compose the reading network are consistently reported to be hypo- or hyperactivated in individuals with dyslexia. When individuals with dyslexia are presented with orthographic stimuli, the visual word form area shows decreased activity when compared to typically reading individuals (Maisog, Einbinder, Flowers, Turkeltaub, & Eden, 2008; Richlan, 2012). In addition, also the temporo-parietal region appears to be consistently hypo-activated in both children (Norton, Beach, & Gabrieli, 2015; Shaywitz et al., 2002) and adults with dyslexia (Paulesu et al., 1996; Rumsey, 1992; Rumsey et al., 1999). Moreover, effective remediation of dyslexia is associated with a growth in activation in this particular area (Eden et al., 2004), suggesting a direct relation between temporo-parietal brain areas and reading. In contrast to the posterior key systems in dyslexia, the (bilateral) inferior frontal system is also considered to be hyperactivated in dyslexia, which is hypothesized to be a compensatory mechanism for the failure of the posterior systems (Brunswick, McCrory, Price, Frith, & Frith, 1999; Shaywitz et al., 2002). Some studies suggest that dyslexia is characterized by compensatory right hemispheric activation during reading (Pugh et al., 2001), although it is also possible that the right hemisphere was never disengaged in early reading development. In addition to several findings of hypo- and hyper-activation in these key regions, functional connectivity between these regions may also related to reading skill (Horwitz, Rumsey, & Donohue, 1998; Koyama et al., 2011). Lastly, dyslexia may also be characterized by impairments in structural, white-matter, connectivity in the arcuate fasciculus and inferior fronto-occipital fasciculus (Beaulieu et al., 2005; Lebel et al., 2013; Vandermosten et al., 2012), the two tracts that connect the three key components of the reading network.

It is difficult to say whether these neurobiological differences are a cause or a consequence of dyslexia. As the reading experience of individuals with dyslexia is, on average, diminished compared to the reading experience of typical readers, reduced

brain activation could hence be the result of less exposure to print stimuli and print to speech conversions (Hoeft et al., 2006). However, some studies compared brain activation of individuals with dyslexia with two separate control groups; one matched on age and one matched on reading level or task performance in the scanner, usually with less or comparable print exposure. The results of these studies also show reduced activation during reading for individuals with dyslexia in left temporo-parietal and fusiform systems, suggesting that these hypo-activations are causally involved in the etiology of dyslexia (Hoeft et al., 2006). Another line of evidence that neurobiological differences are causally involved in reading development are studies that scanned children prior to formal literacy instruction and compared these measures between prereaders with and without an increased risk for dyslexia or compared them with later reading abilities. Functional activation impairments have also been found in temporo-parietal and inferior temporo-occipital regions and compensatory recruitment in the anterior cingulate cortex in prereaders with an increased risk of dyslexia (Specht et al., 2009; Yamada et al., 2011). Additionally, structural deficits in the arcuate fasciculus and inferior fronto-occipital fasciculus have also been reported in prereaders at risk of dyslexia (Vandermosten et al., 2015).

THE PRESENT DISSERTATION

Findings from behavioral and neural theories in dyslexia research point to deficiencies in the quality of (or access to) phonological representations in individuals with dyslexia, although the nature and underlying cause of these deficits is debated. Phonological representations are often measured by means of phonological awareness, speech perception, and less frequently, speech production skills. However, neurocomputational models of speech production suggest that the development and maintenance of adequate phonological representations crucially hinges on the interaction between speech perception and production (Guenther et al., 2006; Hickok, 2012; Houde & Nagarajan, 2011). More specifically, speech feedforward mechanisms map the phonological representation onto motor effectors, and speech sensory mechanisms detect whether the produced sound matches with the auditory and somatosensory targets associated with that phonological representation. Mismatches between the intended and produced sound are detected and used to correct speech mistakes and, when consistently encountering an error, update the motor and/or sensory traces associated with phonological representations (Guenther et al., 2006). This monitoring of self-produced speech and using the resulting information to alter speech production is called the sensorimotor control of speech. Probing this sensorimotor control of

speech, and examining how it relates to reading skill opens a new window to examine phonological deficits in dyslexia and reading development and this will be the central topic of the current dissertation.

Administering online alterations in auditory feedback induces a mismatch between intended and produced sounds and is a frequently used paradigm to examine the sensorimotor control of speech. Humans are hypothesized to use auditory goals in their motor planning of speech (Villacorta, Perkell, & Guenther, 2007). Measuring changes in ones' speech production under conditions of altered auditory feedback provides information on (i) an individual's auditory target and (ii) the ability to use auditory feedback to change speech production in order to reach the auditory goal (Guenther, 2015; Niziolek & Guenther, 2013). A few studies applied alterations in auditory feedback in individuals with and without dyslexia, showing that reading accuracy and fluency was relatively less impaired in individuals with dyslexia under conditions of masked (Breznitz, 1997) or pitch-shifted (Carter, Rastatter, Walker, & O'Brien, 2009; Rastatter, Barrow, & Stuart, 2007) altered feedback. Although these studies suggest individuals with dyslexia process auditory feedback differently, the methods of altered feedback are not specific to individual phonological representations and this makes it harder to relate these studies to the observed phonological deficiencies in dyslexia. Manipulating individual phonemes on a trial-by-trial basis, while measuring subsequent speech productions, could be seen as more promising since it allows to examine the dynamics of phonological representations (MacDonald, Johnson, Forsythe, Plante, & Munhall, 2012; Villacorta et al., 2007).

One way to examine the sensorimotor control of individual phonemes is by studying formant adaptations. Formant adaptation studies are studies in which a formant—a burst in the speech signal—is altered. Each vowel is characterized by relatively fixed formant values and changing a formant can lead one vowel to sound like another. Experiments that applied this paradigm usually consist of several phases: a baseline phase, in which the formants are not altered and in which the participant can get used to the paradigm and to his own auditory feedback; a ramp phase, in which a formant is gradually altered to avoid conscious awareness of the manipulation; a hold phase, in which the alteration of the formant is kept at maximal for a number of trials; and an after-effect phase in which the feedback is back to normal and in which participants can return to their 'baseline' formant production of that vowel. The typical response of participants is to change their formant production in the opposite direction of the altered auditory signal (Burnett, Senner, & Larson, 1997; Lametti, Nasir, & Ostry, 2012; Niziolek & Guenther, 2013; Villacorta et al., 2007). An advantage of using a formant

adaptation paradigm is that participants are usually not aware of the manipulation and the response to altered feedback appears to occur automatically and without effort (Tourville, Reilly, & Guenther, 2008).

Aims and research questions

The aim of the present dissertation is to examine the role of sensorimotor control of speech in differences in reading skill. Measuring the sensorimotor control of speech provides a direct insight in the underlying mechanism that is hypothesized to be causally involved in the development of phonological representations, as well as insight in the quality of phonological representations themselves. A core hypothesis of dyslexia is that deficient phonological representations hamper the stabilization and automatization of grapheme-phoneme correspondences. The current dissertation describes the first studies that used measures of the sensorimotor control of speech and relates them to reading skill and reading development. More specifically, the sensorimotor control of individual's speech sounds (i.e. phonemes) is examined. The following research questions are addressed in the present dissertation:

1. Can dyslexia be characterized by an impaired sensorimotor control of speech?
2. Is there also neurobiological evidence for such impaired sensorimotor control of speech?
3. Are individual differences in reading problems associated with sensorimotor control of speech?
4. How are changes in reading skill over time associated with sensorimotor control of speech?

Answering these research questions will provide a new perspective on theories of dyslexia. First, since the sensorimotor control of speech is usually effortless and occurs automatically, the results of these studies can indicate whether the phonological representations themselves are impaired in dyslexia, or that individuals with dyslexia have difficulties in accessing phonological representations. Second, the sensorimotor control of phonemes, under conditions of systematic alterations in auditory feedback, is sensitive to statistical learning. The speed of adaptation in individuals with dyslexia could provide information on how quickly individuals with dyslexia extract, and respond to, statistical (ir)regularities in the speech signal. Lastly, if dyslexia is indeed associated with impaired sensorimotor control of phonemes, a wide range of possibilities for follow up studies exists to distinguish between speech specific and auditory/sensory/cognitive theories on dyslexia.

OUTLINE OF THE DISSERTATION

The aim of the thesis was to examine whether and how the sensorimotor control of speech is related to reading skill and reading development.

In Chapter 2, I examined whether adults with dyslexia show a deficit in sensorimotor control of speech**.** First, we examined whether a similar number individuals with and without dyslexia showed the typical adaptation pattern, by opposing their speech production to the perturbation in auditory feedback and whether the baseline formant production and variability were similar across groups. Second, we compared the response to altered auditory feedback experiment, in which the first formant of the /ɛ/ vowel was increased with 25-30%, between adults with and without dyslexia. Lastly, we examined whether individual differences, associated with their response to altered auditory feedback, in participants with dyslexia were related to their reading and reading-related task performance.

In Chapter 3, I measured whether dyslexia is characterized by deficient neural processing in brain areas and structures that are hypothesized to support the sensorimotor control of speech. First, using structural MRI, I examined whether the fractional anisotropy—a measure of white matter organization—of the arcuate fasciculus was lower in adults with dyslexia, compared to typically reading adults. The arcuate fasciculus structurally connects superior temporal regions (involved in speech perception) with inferior frontal areas (involved in speech production) (Catani & de Schotten, 2008). Second, using functional MRI, I investigated the efficiency of a large speech network. Efficient communication between speech production and speech sensory areas is required in order to optimally use sensory feedback to adapt speech production. Third, I examined the functional activation patterns during speech perception and production to test whether speech sensory areas are activated during speech production and vice versa.

Chapter 4 investigates whether individual differences in reading and reading-related abilities in children with dyslexia can be accounted for by their sensorimotor control of speech, and whether the fractional anisotropy of the arcuate fasciculus was related to the response to altered feedback. To force all participants to oppose their speech production in response to the altered feedback, I measured each individuals /i/ and /ɛ/ sounds and applied a complete /i/ to /ɛ/ change in the altered auditory feedback experiment. First, in an attempt to replicate the results of Chapter 2, I compared the response to altered auditory feedback between children with and without dyslexia. Second, for children with dyslexia only, I examined whether individual differences in reading skill, response to intervention, rapid naming and phonological awareness were

related to the response to altered auditory feedback. Lastly, the role of the arcuate fasciculus, and its interaction with behavioral measures, in the response to altered feedback was examined.

In Chapter 5, I examined whether the response to altered auditory feedback was related to different stages of reading development, and whether it was associated with important precursors of reading development. Moreover, I did so in two contrastive orthographies with respect to the orthographic depth. Some studies propose that the role of phonological representations and reading skill is dependent on orthographic transparency (Caravolas, Volín, & Hulme, 2005; Ziegler et al., 2010). Transparent orthographies contain mappings between spelling and pronunciations that are regular or consistent, whereas opaque orthographies have multivalent or 'inconsistent' mappings between spelling and pronunciations (Borgwaldt, Hellwig, & De Groot, 2005). In this study, more than 200 American and Dutch children between 4-8 years old, participated in an altered auditory feedback paradigm. First, I examined whether being able to read is related to the response to altered auditory feedback. Second, I investigated whether this was dependent on reading English or Dutch. Third, I examined whether the response to altered auditory feedback was related to precursors of reading (phonological awareness, rapid naming, letter knowledge), separately for the preliterate and literate children.

Chapter 6 discusses the finding of the present thesis. I will first propose a new perspective on the origin of dyslexia that captures the results of the presented studies. Second, I will discuss what the results of the presented studies imply for several theories of dyslexia. Lastly, I will discuss whether deficiencies in sensorimotor control of speech are a cause or a consequence of dyslexia. Each section will be concluded by follow-up suggestions for future research.

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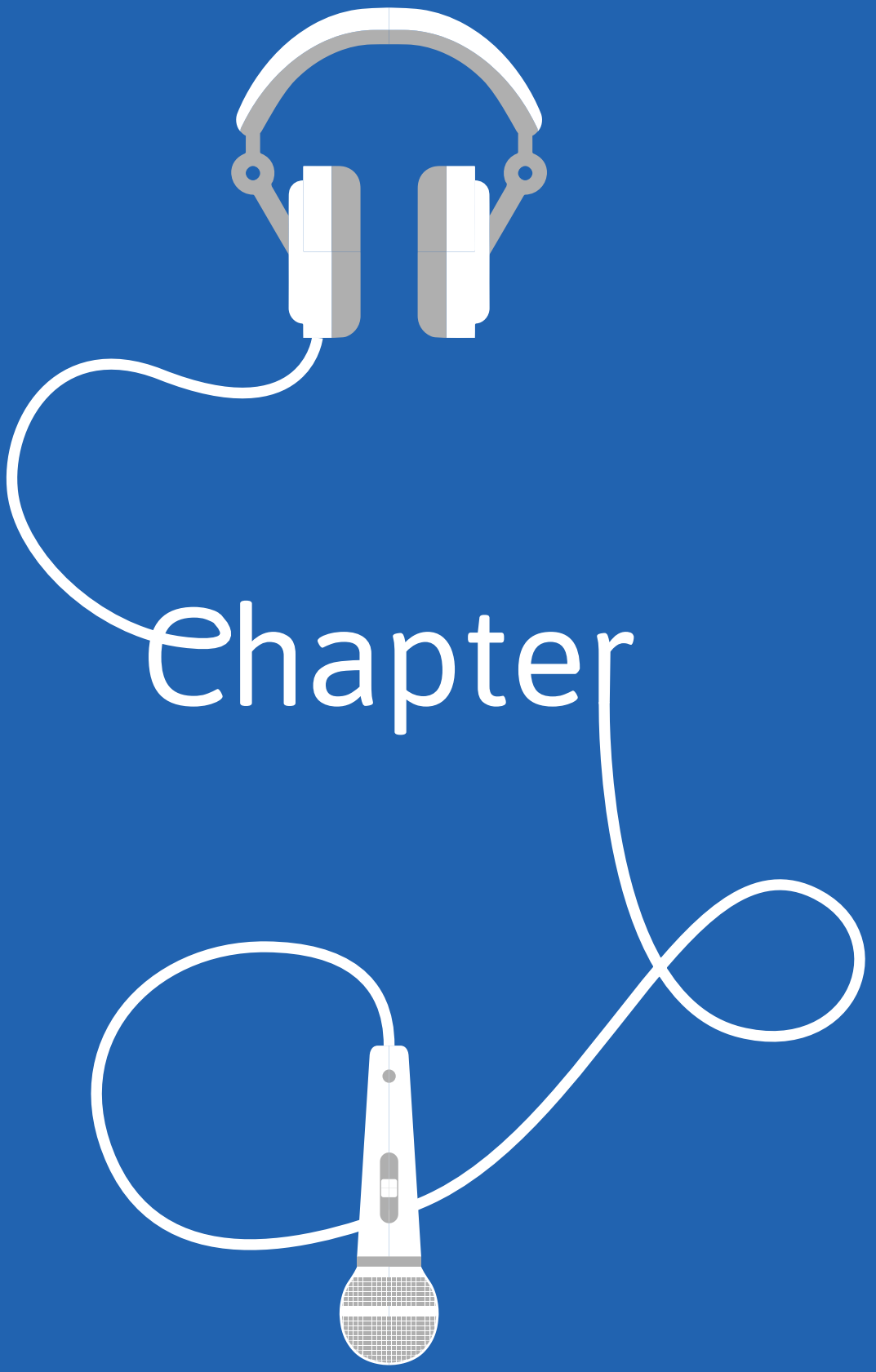
Chapter

INCREASED RESPONSE TO ALTERED AUDITORY FEEDBACK IN DYSLEXIA: A WEAKER SENSORIMOTOR MAGNET IMPLIED IN THE PHONOLOGICAL DEFICIT

M.R. van den Bunt[1*]
M.A. Groen[1]
T. Ito[2,4,5]
A.A. Francisco[1]
V.L. Gracco[2,3]
K.R. Pugh[2]
L. Verhoeven[1]

[1] Behavioural Science Institute, Radboud University, Nijmegen, The Netherlands
[2] Haskins Laboratories, Yale University, CT, New Haven, USA
[3] Centre for Research on Brain, Language & Music, McGill University, Montreal, Canada
[4] Université Grenoble Alpes, GIPSA-Lab, Grenoble, France
[5] CNRS, GIPSA-Lab, Grenoble, France
[*] Corresponding author: Mark van den Bunt; m.vandenbunt@pwo.ru.nl; phone nr.: +31243612430; Montessorilaan 3, 6525 HR Nijmegen, The Netherlands

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ABSTRACT

Developmental dyslexia is characterized by a deficit in phonological representations, but the nature of this deficit is currently debated. Neurocomputational models suggest that the quality of phonological representations depends on the integrity of speech sensory and motor feed-forward and feedback mechanisms. In this study, we examined these mechanisms in adults with dyslexia and typically-reading controls, by investigating their response to altered version of the first formants of their own speech productions, fed back in real-time. Linear mixed-effects modeling showed that adults with dyslexia adapted more strongly in response to altered feedback and returned to baseline more weakly when feedback was unaltered. The results of the current study are consistent with the notion that the phonological deficit in DD is associated with a 'weaker' magnet for phonological representations.

INTRODUCTION

Developmental dyslexia (DD)—with a prevalence estimate of approximately seven percent across languages a relatively common condition (Goswami, 2015)—is defined as a brain-based difficulty in acquiring fluent word decoding skills (Lyon, Shaywitz, & Shaywitz, 2003) and interferes considerably with level and amount of educational and occupational activities (Kurtner et al., 2007). Despite adequate general cognitive abilities and appropriate educational opportunities, individuals with DD fail to automate the associations between graphemes and phonemes (Liberman, Shankweiler, & Liberman, 1989). There is considerable consensus that deficits in the development of phonological processing, and more specifically, the quality of and/or access to phonological representations, are implicated in DD (Boada & Pennington, 2006; Boets et al., 2013; Sprugevica & Høien, 2003). Notably, these phonological deficits are reported to largely persist into adulthood (Shaywitz et al., 1999; Wilson & Lesaux, 2001). To date, research on phonological abilities in DD has focused predominantly on either speech perception (e.g., Ziegler, Pech-Georgel, George, & Lorenzi, 2009) or production (e.g., Foy & Mann, 2012). However, current models of speech production suggest that the interaction between speech perception and production might be crucial in understanding the development of phonological representations (Guenther, Ghosh, & Tourville, 2006; Hickok, Houde, & Rong, 2011). Indeed, poorer performance by individuals with dyslexia on non-word repetition tasks (Messbauer & de Jong, 2003)—an integrated measure of speech perception and production—can be seen as an indication that perception-production interaction might be deficient in developmental dyslexia (Coady & Evans, 2008; Snowling, Chiat, & Hulme, 1991). As such, we propose that studying speech perception and production interaction is important in understanding the nature of the phonological deficit in developmental dyslexia, and do so in the current study by assessing online adjustments of speech production following altered auditory feedback.

A phonological processing deficit in developmental dyslexia

The evidence for phonological processing deficits in DD comes from a vast number of studies starting in the 1980's (Katz, 1986; Snowling, 1981). In more recent studies, phonological awareness and rapid naming are marked as important predictors of reading acquisition (Thompson et al., 2015; Van der Leij et al., 2013), although the strength of these predictors is reported to vary across orthographies (Caravolas, Lervåg, Defior, Seidlová Málková, & Hulme, 2013; Georgiou, Parrila, & Papadopoulos, 2008; Ziegler et al., 2010; but see Vaessen et al., 2010). Phonological awareness appears to develop on a continuum from being able to segment syllables and detect rhyme to individual sound segmentation and manipulation (Anthony & Francis, 2005). According

to the segmentation theory, the quality of phonological representations is dependent on this development, and deficient segmentation is associated with reduced reading ability (Metsala & Walley, 1998). In contrast, Elbro and colleagues suggested that a lack in distinctness of these representations underlies the phonological deficit in DD (Elbro, Borstrom, & Petersen, 1998; Elbro, 1996, 1998). The deficient phonological representations have been described by various classifications such as immature, underspecified, fuzzy, fragile, non-robust and indistinct, which indicates that the exact nature of the deficit in phonological representations is rather vague (e.g., Boada & Pennington, 2006). Adequate processing of speech input, as well as articulatory output representations, are implied in the development of phonological representations (Nittrouer, 1996). Consequently, it is often assumed that poor speech perception and production skills underlie the impaired phonological skills in DD (Foy & Mann, 2001, 2012; Mann & Foy, 2007; Preston & Edwards, 2010).

People with DD indeed show deficient abilities in speech perception and production. Many studies on speech perception point to reduced abilities to identify and discriminate between phonemes, both in optimal (De Weirdt, 1988; Godfrey, Syrdal-Lasky, Millay, & Knox, 1981) and in adverse listening conditions (Ziegler, Pech-Georgel, George, Alario, & Lorenzi, 2005; Ziegler et al., 2009). Although many studies focused on deficiencies in perceiving consonants, the perception and production of vowels is also less precisely defined (Bertucci, Hook, Haynes, Macaruso, & Bickley, 2003; Stark & Heinz, 1996). Although a large number of studies showed perception deficits in DD, it should be noted that speech perception deficits were not always found in the majority of people with DD (Manis et al., 1997), not for all phonetic contrasts (Cornelissen, Hansen, Bradley, & Stein, 1996), and not always in silent (Ziegler et al., 2009) or in noise conditions (Hazan, Messaoud-Galusi, & Rosen, 2012; Law, Vandermosten, Ghesquiere, & Wouters, 2014). Although the majority of studies show that people with DD have deficient perceptual abilities, resulting in less precise or degraded phonological representations, another perspective on the perception 'deficit' comes from Serniclaes, Van Heghe, Mousty, Carré, and Sprenger-Charolles (2004). These authors provided evidence that people with DD remain sensitive to allophonic variants within phoneme categories, hence hindering phoneme-level representations to develop adequately (but see Ramus & Szenkovits, 2008). This theory suggests that speech perception is not degraded in DD on an acoustic level, but instead, not adequately attuned to the phonetic contrasts present in the native language. This hypothesis has further been supported using behavioral and neuroimaging measures in children at-risk for DD (Noordenbos, Segers, Serniclaes, Mitterer, & Verhoeven, 2012a, 2012b). With regard to speech production, it has been

shown that both articulatory skills (Catts, 1986, 1989) and oral motor skills are impaired in people with DD (Malek, Amiri, Hekmati, Pirzadeh, & Gholizadeh, 2013; Smith, Roberts, Lambrecht-Smith, Locke, & Bennett, 2006).

The nature of these phonological deficits is, however, challenged by a series of experiments by Ramus and Szenkovits (2008), suggesting that the phonological deficit is related to the access to, rather than the quality of, phonological representations. The authors claim that the phonological deficit becomes particularly apparent when tasks place strong demands on short-term memory, conscious awareness and speed, which impedes fluent retrieval, extraction and manipulation of phonological representations. The same study indicates that individuals with DD are equally unable to discriminate between foreign speech sounds, and hence questions the theory of an allophonic mode of perception in DD (posed by Serniclaes et al., 2004). Additionally, neuroimaging findings from Boets and colleagues (2013) report impaired connectivity between frontal and temporal language areas, which hampers 'efficient access to otherwise intact representations of speech sounds' (p.1254). These studies do not reject a phonological deficit in DD, but suggest an alternative formulation of the impairment.

A frequently employed measure in the DD literature that provides an integrated, but non-decomposable, measure of speech perception and production is non-word repetition. Many consider non-word repetition to be primarily a measure of phonological short-term memory (e.g., Ramachandra, Hewitt, & Brackenbury, 2011), however, multiple processes are involved and reflected by non-word repetition. Each of these processes, including auditory processing, phonological processing, phonological storage, speech-motor planning and speech output (Gathercole, 2006), were found to be related to DD. It can be assumed that poor or inaccessible phonological representations constrain the ability to adequately process the auditory input and produce the auditory output in non-word repetition tasks. Indeed, deficient ability to repeat non-words has been reported in a variety of language disorders (e.g., stuttering (Sasisekaran, 2013) and specific-language impairment (Edwards & Lahey, 1998)) and has consistently been shown in people with, or at risk for, DD (Catts, Adlof, Hogan, & Weismer, 2005; de Bree, Rispens, & Gerrits, 2007). A recent meta-analysis of the role of non-word repetition in DD also concluded that people with DD perform reliably worse on non-word repetition tasks, with large effect sizes when compared to chronological age matched controls and small to moderate effects when compared to reading level matched controls (Melby-Lervåg & Lervåg, 2012).

Current models of speech production suggest that it is not either speech perception or production that is important in the adequate development of phonological representations but that the interaction between perception and production is crucial for this development (Hickok, 2012; Houde & Nagarajan, 2011; Tourville & Guenther, 2011). Speech perception and production have largely been investigated separately in DD, but probing how perception and production interact might be vital in understanding the nature of the phonological deficit in DD. Attempts to study speech perception-production interaction have been made by administering a non-word repetition task in the context of a paired associate learning task, in which the same non-word had to be repeated multiple times. People with DD have been reported to acquire new phonological forms more slowly (Messbauer, & De Jong, 2003), particularly in case of phonologically complex non-words (Mayringer, & Wimmer, 2000). These deficiencies in non-word learning in people with DD could be due to impairments in speech perception-production interactions, but they do not speak to the mechanism(s) underlying such a deficient interaction. In contrast, studies outside the DD literature have provided testable models about the formation and modulation of phonological representations (Guenther et al., 2006; Hickok et al., 2011) and applying these models to a DD population could potentially help to explain the nature of the phonological deficit.

Phonological representations and the role of altered auditory feedback

Neurocomputational models have indicated that the quality of phonological representations depends on the integrity of speech sensory and speech motor feed-forward and feedback mechanisms (Hickok, 2012; Houde & Nagarajan, 2011; Tourville & Guenther, 2011). Two prominent and neurally plausible theories on how phonological representations are formed and adjusted are the Directions Into Velocities of Articulators (DIVA; Guenther et al., 2006) and State-Feedback Control (SFC, Houde & Nagarajan, 2011) models. Although these models differ on some fundamental issues (e.g., whether the dynamics of the articulators are fully taken into account, for more discussion see Houde & Nagarajan, 2011; Riley-Graham, 2011; Tourville & Guenther, 2011), both models adopt a feed-forward trace that maps phonological representations to motor effectors, and a feedback trace that controls whether the sensory consequences of the speech realization match with the predicted sensory consequences. Mismatches are used to adjust the phonological representation. Once adequate feed-forward commands are formed, the inefficient and slow feedback system becomes redundant and will largely disengage (Guenther et al., 2006). Perturbations in auditory feedback induce a conflict between these motoric and sensory traces associated with phonological representations. Villacorta and colleagues (2007) hypothesized that humans use auditory goals in their motor planning. By measuring changes in an individual's speech productions under

conditions of altered auditory feedback, we acquire information about an individual's auditory target associated with a particular phonological representation, and his ability to adjust his speech production to match those auditory goals (Guenther, 2015; Niziolek & Guenther, 2013). As such this paradigm enables us to quantify aspects of phonological representations.

The presence and quality of auditory feedback during development has indeed been shown to significantly affect skills in speech production. For instance, there is evidence that pre-lingual deaf children have problems in developing intelligible speech skills (Oller & Eilers, 1988) and that speech production of children with cochlear implants, who receive better auditory input, is often more adequate than that of children with strong hearing loss using hearing aids (Baudonck, Dhooge, D'haeseleer, & Van Lierde, 2010). Several studies have shown that delaying auditory feedback or masking auditory feedback by noise, affects speech production both in typical (Amazi & Garber, 1982; Chon, Kraft, Zhang, Loucks, & Ambrose, 2012; Sasisekaran, 2012) and clinical populations (Hudock & Kalinowski, 2014).

With regard to DD, reading under conditions in which auditory feedback was masked (by playing familiar tunes over headphones; Breznitz, 1997) or in which the participants' pitch was shifted (Carter, Rastatter, Walker, & O'Brien, 2009; Rastatter, Barrow, & Stuart, 2007) significantly increased reading accuracy, fluency and comprehension in both children and adults with DD. Although these studies show that people with DD process auditory feedback differently—which apparently impedes reading—they are not informative as to the mechanism behind this difference. Manipulating auditory feedback on a trial-by-trial basis can be seen as promising in this respect since it allows us to examine how auditory feedback is implicated in adjusting phonological representations dynamically (MacDonald, Johnson, Forsythe, Plante, & Munhall, 2012; Villacorta, Perkell, & Guenther, 2007).

Studies in which formants spectral peaks in the sound system are manipulated and fed-back in real-time may provide better insight into the mechanism(s) of adjusting phonological representations. In classical studies it has been found that formants largely determine the identity of vowels, and that manipulating the first formant can cause one vowel to sound like another (Delattre, Liberman, Cooper, & Gerstman, 1952). Formant adaptation studies generally consist of a baseline phase in which the normal distribution of the participants' formant production is measured. This phase is followed by a ramp and hold phase in which one or more formants are gradually adapted (either increased or decreased) over trials and fed back in real-time. The last phase consists of trials without altered feedback to measure whether the participants' response

returned to baseline. It has been found that participants usually adapt to the auditory perturbation by shifting their formant production in the opposite direction of the manipulation (Houde & Jordan, 1998, 2002; MacDonald et al., 2012; Purcell & Munhall, 2006; Villacorta et al., 2007). There is evidence that participants are able to modify their response to correct for multiple auditory transformations and that the modification of a phoneme tends to generalize across different words (Rochet-Capellan & Ostry, 2011). These production changes are strong enough to be partly retained when feedback is blocked by noise (Houde & Jordan, 2002). When feedback is back to normal, the return to baseline was found to be gradual and not dependent on the number of trials of maximal perturbation (Purcell & Munhall, 2006; Villacorta et al., 2007). Another important issue related to the amount of adaptation is whether the auditory perturbation changes the phoneme identity or varies only on a sub-phonemic level. Near and across- phoneme boundary perturbation has been reported to result in stronger adaptation (Niziolek & Guenther, 2013). Niziolek and Guenther (2013) report these effects in the context of the Native Language Magnet theory (also known as the perceptual magnet theory; Kuhl, 1991; Feldman, Griffiths, & Morgan, 2009). Its core claim is that a phonetic category prototype functions as an attractor (i.e. magnet) that warps the psychoacoustic space, resulting in poorer discriminability for neighboring stimuli near the category prototype (i.e. a narrower space), and better discriminability farther away from the prototype (i.e. a stretched space). Perturbations in the auditory signal are hence expected to elicit a stronger response when the presented auditory stimulus is farther away from the phonemic category prototype.

It should be noted however, that within and across studies, the amount of adaptation varies widely across individuals and several accounts exist for this variability. For instance, Burnett and colleagues (1997; 1998) reported that some participants changed their formant production in the direction of the adaptation. It was suggested that these participants might use an external auditory reference for adequate formant frequencies, rather than an internally set reference (Burnett, 1998). Alternatively, Lametti, Nasir and Ostry (2012) show that individuals differ in their preferential reliance on auditory or somatosensory feedback, which could well explain why some individuals do and some do not adapt under conditions of altered auditory feedback. This variability in response is certainly not unique to speech perception-production interaction. For instance the McGurk effect, a traditional measure of audiovisual integration and well known in the DD field, shows a dramatic diversity of responses both across individuals and across used stimuli (Basu Mallick, Magnotti, & Beauchamp, 2015).

The aforementioned studies show how auditory feedback affects speech production in the typical population. Studies on these formant adaptation effects in clinical disorders are scarce. Using simulations, Civier, Tasko and Guenther (2010) showed that stuttering may be caused by deficits in speech feed-forward and feedback mechanisms (as explicated in the DIVA model). Their study suggests that stutterers rely too heavily on the auditory feedback trace to control speech and hence are more sensitive to changes in auditory feedback. Another relevant clinical group concerns specific language impairment (SLI) which shares many characteristics with DD, including a phonological processing deficit (Bishop & Snowling, 2004; Edwards & Lahey, 1998). In a small case-control comparison, it was found that children with SLI showed more adaptation than their typically developing peers when the frequency of the first formant of a vowel was altered, and furthermore, did not fully return to baseline when feedback was back to normal (Holmes, 2012). To date, however, no attempt has been made to examine whether DD is characterized by differences in response to this altered auditory feedback in order to shed light on the nature of the phonological deficit.

Present study

The present study aimed to gain better insight into the nature of the phonological deficit in DD by examining the ability to modulate existing phonological representations in adults with developmental dyslexia (DD) and typically reading controls (TR). If the phonological deficit in DD is characterized by deficiencies in speech perception-production interaction, individuals with DD should adjust their productions differently when auditory feedback (i.e., speech perception) is manipulated.

To assess these speech feed-forward and feedback mechanisms, we used an altered auditory feedback design (Houde & Jordan, 1998; Purcell & Munhall, 2006; Rochet-Capellan & Ostry, 2011) changing the perception of the first formant of the vowel in the participants production of the word /bɛp/. After a first phase, during which the first formant of the /ɛ/ was not altered (i.e. baseline), the frequency of the first formant gradually was increased during the second phase (ramp), and was held at maximal perturbation in the third phase (hold). Lastly, the manipulation was switched off and the frequency of the first formant fed back to the participant was unchanged from their production for the last phase of the experiment (after-effect). Typically, participants will respond to the manipulation by adjusting the frequency of the first formant of their productions in the opposite direction. Changes in formant production in response to alterations in auditory feedback are indicative of how perceiving auditory manipulations interacts with producing speech. Differences in the amount of adaptation between individuals with DD and typical readers could be caused by several different factors. For

instance, both stronger and weaker adaptation during the ramp phase could be purely related to perceptual deficiencies in DD (allophonic perception or degraded perception, respectively), but also to motor impairments (e.g. unstable or rigid motor commands). The pattern of the responses to the different phases of feedback, however, could clarify whether DD is characterized best by a perceptual deficit, a motor deficit, or a deficient interaction between perception and production.

The hypothesized overreliance on auditory feedback in stuttering (Civier et al., 2010), as well as the stronger response to altered feedback in SLI (Holmes, 2012), led us to expect a stronger response to altered auditory feedback in adults with DD during the ramp and hold phase. This would also be in line with the allophonic mode of perception in DD proposed by Serniclaes and colleagues (Serniclaes et al., 2004). In contrast, a reduced response to altered feedback in the ramp and hold phase, we think, would be consistent with the 'phonological access deficit' (Boets et al., 2013; Ramus & Szenkovits, 2008). The expectations for the after-effect phase were harder to explicate on these accounts. If DD is indeed characterized by an overreliance on auditory feedback and/or an allophonic perception mode, a stronger return to baseline might be expected. However, children with SLI did not return to baseline to the same extent as typically developing controls (Holmes, 2012), and there may be parallels in DD. This pattern of responses would fit with a more general hypothesis predicated on the notion that, irrespective of the mechanism, phonological representations in DD are of lower quality and may act as 'weaker' attractors (Anderson, Morgan, & White, 2003; Baker, Trofimovich, Mack, & Flege, 2002) or perceptual magnets (Iverson & Kuhl, 2000; Kuhl, 1991). On that notion we might expect stronger adaptation and weaker de-adaptation in DD. In addition to examining these group responses, we explored whether individual differences in the response to altered auditory feedback were associated with phonological and reading abilities. Given our assumption that this response taps into aspects of phonological representations that are relevant to reading, we expected this to be the case.

METHODS

Participants

Twenty typically reading (TR) university students (14 women: M_{age} = 22.32 years; SD_{age} = 2.7 years) and 22 university students with developmental dyslexia (DD) (17 women; M_{age} = 23.13 years; SD_{age} = 2.7 years) were included in this study. All participants were native speakers of Dutch. Participants were approached via email as they took part in earlier studies in our lab and had consented to be contacted in this manner. As part

of these earlier studies participants were recently (<12 months) characterized in terms of reading and phonological awareness. All participants received course credits or a monetary compensation for participation.

To be included in the DD group, participants had to be officially diagnosed and perform below the 30th percentile on reading accuracy or below the 30th percentile on reading time. TR students were required to perform above these thresholds. Additionally, all participants passed the hearing screening, perceiving pure tones presented at less than 30 dB at 250 Hz, 500 kHz, 1 kHz, 2 kHz, and 4 kHz in both ears. One participant was excluded due to a cold, which significantly affected speech production. Participant characteristics are provided in Table 1.

TABLE 1. Participant characteristics for the readers with developmental dyslexia (DD) and the typically reading controls (TR)

	TR (N = 19)		DD (N = 22)		Significance
	M	SD	M	SD	t / *U* test
Reading (errors)	8.63	4.46	16.68	7.05	t=4.288**
Reading (time)^	243.74	8.99	303.41	27.90	*U*=2.0**
Phonological awareness (accuracy)	16.89	2.16	17.18	1.99	t=-.443
Phonological awareness (time)^	97.58	17.64	123.68	32.79	*U*=87.5*

^Mann-Whitney U test as the distribution in the DD group was non-parametrical
*p < .01, **p < .001

Materials

Reading

In order to assess reading ability, all participants were asked to read aloud a 582-word text, a subtest of a standardized Dutch test battery for the diagnosis of DD in adolescents and adults (GL&Schr – *Test voor gevorderd Lezen en Schrijven* [De Pessemier & Andries, 2009]). Guttman split-half reliability for reading accuracy and reading time was adequate (respectively .77 and .90). The text was divided into paragraphs and the number of phonologically complex and unfamiliar words increased for each paragraph to evoke reading errors. The produced reading was recorded to optimize scoring accuracy. Omissions, additions, replacements and inversions were counted as errors, and were carefully determined by listening to the recorded audiofiles. Norm scores from the manual were used to calculate percentiles in order to determine whether the participants fulfilled the inclusion criteria. The raw test scores for the number of errors and the time to complete the task (in seconds) were used for all statistical analyses.

Phonological awareness

Phonological awareness was measured using the phonological reversal task from the same test battery (GL&Schr – *Test voor gevorderd Lezen en Schrijven* [De Pessemier & Andries, 2009]). Reliability of this task had been calculated at r = .90 (Guttman split-half reliability). In each trial, two audio-recorded items were presented to the participant who was asked to indicate (yes-no) whether the second item was the phonological reverse of the first. A next trial was started after the experimenter pressed the button to continue. The task started with six practice trials during which feedback was provided. The experimental part consisted of 20 items for which accuracy per item and total duration for all items was registered. Total number correct (accuracy) and the time to complete the task (in seconds) are reported.

Altered Auditory Feedback task

In the altered auditory feedback task, participants were asked to produce the word / bɛp/ when a specific blue cartoon figure popped up on the screen. In case of other cartoon figures, participants were asked to remain silent. The blue figure appeared in ± 70% of the presentations and speech was automatically recorded for two seconds to capture the /bɛp/ production. Participants were explicitly instructed to say nothing else than /bɛp/ until the experiment was finished. A total number of 95 productions were collected for each participant. The participants' speech production was manipulated and fed-back in real time with approximately 10 ms of delay.

To ensure that the participants perceived the altered signal instead of their own voice, the speech signal was amplified and accompanied by 70dB of pink noise. The pink noise further reduced the perception of the air- and bone conduction of the produced signal. The experiment consisted of four phases. The first phase (baseline) consisted of 30 trials during which feedback was not altered. The second phase (ramp) consisted of 25 trials during which frequency of the first formant (F1) of the speech production was gradually and imperceptibly shifted until a maximal increase of approximately 30% was reached. The third phase (hold) consisted of 25 trials during which the F1 was maximally altered. The last phase (after-effect) consisted of 15 trials during which the altered feedback was completely shut off.

Equipment: A microphone (e835 FX; Sennheiser electronic GmbH & Co. KG, Wedemark, Germany) was placed in close proximity to the mouth and participants wore a headphone (HD360 Pro, Sennheiser electronic GmbH & Co. KG, Wedemark, Germany). The produced speech was amplified using the microphone preamplifier (Tube UltraGain MIC100 , Behringer GmbH, Kirchardt, Germany) and split into two streams. In one stream, the F1 was altered by a sound signal processor (VoiceOne; TC Helicon Vocal Technologies,

Victoria, BC, Canada). The signal in the other stream was unaltered. Using analog filters (852, Wavetek, San Deigo, CA, USA), we applied a low pass filter on the altered stream (<1 kHz; F1 of /ɛ/ is below 1 kHz) and a high pass filter (>1 kHz) on the unaltered stream. The sound signal processor was controlled via midi in an external audio device (Roland UA-25 EX, Hamamatsu, Japan). As the sound signal processor takes approximately 10 milliseconds to alter the signal, the high frequency stream was delayed by 10ms using an audio delay box (DataVideo AD100; Datavideo Technologies Europe BV; Utrecht; The Netherlands). Finally, the two streams and the noise signal were mixed (Skytec STM3004, Skytronic Ltd, Manchester, UK) and amplified through a headphone amplifier (HA400, Behringer GmbH, Kirchardt, Germany).

Procedure

Informed consent was signed after the participant arrived at the lab. Participants were then positioned in front of the microphone and a monitor. Subsequently, the amplification of the signal was increased as much as was still comfortable for the participant. The noise volume always remained at 70dB. All participants started with a 15-trial practice block and when everything was clear, progressed to the experimental items. After the experiment, participants were asked whether they noted anything special during the experiment, and if not, more specifically, whether they noted anything remarkable about the sound. In case both answers were negative, we explained exactly what we did and asked whether the participant now recognized the manipulation. None of the participants confirmed to be aware of, or recognized the manipulation. Participants left after a short debriefing.

Analysis

Given the considerable variability in the amount of adaption observed across individuals, as well as the assumed fundamentally different underlying mechanisms of the presence or absence of the typical adaptation response (Burnett et al., 1997; Lametti et al., 2012), we first examined whether a similar number of people with DD and TR controls showed the typical adaptation response when the frequency of the first formant was altered. Then, using the data from these adapters only, we asked whether people with DD differed from TR controls in the magnitude of adaptation for the distinct phases of the experiment.

The first formants were calculated using linear predictive coding (Rabiner & Schafer, 1978) in Matlab 2014 (The MathWorks Inc., Natick, MA, USA) after the first author manually indicated the center of each vowel. Outlying formants (>3 standard deviations, calculated per phase) were removed from all analyses. We then determined

for each participant whether the response should be classified as an adapting or a non-adapting response to the feedback alteration. This was determined by comparing the F1 frequency during the hold phase with the baseline phase (one-sided t-test; $\alpha = 0.05$). The typical response is a significant depression of F1 in the produced vowel sound in response to the manipulation of F1 increase. We categorized participants who showed this typical adaptive behavior as adapters. Non-adapters were participants who either did not respond to the perturbation at all (possibly due to relying more heavily on somatosensory feedback [see Lametti et al., 2012]) or followed the manipulation in the same direction (possibly due to 'external auditory goals' [see Burnett et al., 1997;1998]). Independent t-tests analyses were performed to ensure that the frequency and variability of the baseline F1 productions were comparable across the TR and DD groups. Additionally, independent t-test were ran to ensure that behavioral measures (reading and phonological awareness) did not relate to (non-)adapting to the manipulation. Chi-square tests were performed to ensure the number of adapters between groups as well as the gender distribution within groups were comparable. All further analyses were performed on adapters only.

We performed linear mixed effects modeling only using the lmer function of the lme4 package (Bates, Maechler, Bolker, & Walker, 2014) in R (R Core Team, 2014) on the raw F1 scores per feedback phase with gender, trial, phase of feedback and group (i.e. DD vs TR) as fixed factors. Gender was added to the models since formant frequencies are reported to be systematically lower in men (Peterson & Barney, 1952). The feedback phase is entered as a fixed factor, rather than an interval variable, to allow differentiating between the absence of altered feedback in the baseline and after-effect phase. The best model fit was obtained by conduction ANOVA's on sequential models, starting from simple (by entering main effects), gradually moving to complex models (by entering different interaction effects). Standardized F1 scores (by using the average and standard deviation of the baseline phase) were only used for graphical purposes and correlational analyses.

Exploratory correlational analyses were performed to relate individual differences in response to altered auditory feedback to differences in phonological and reading abilities, both within and across groups. Particularly important in studying the robustness of phonological representations, is how fast participants deviate from baseline (calculated using the slope in the ramp phase), how much participants ultimately adjust (calculated using the mean standardized frequency of the first formant during the hold phase), and how fast participants return to baseline (calculated using the slope in the after-effect

phase). The slopes were calculated, for these correlational analyses only, by dividing the difference between the average of the first and last five trials of a phase by the total number of trials in that phase.

RESULTS

Our first research question concerned the number of people showing the typical adaptation response to altered auditory feedback. There was considerable variability in response to the manipulation: A total of 25 (61%) participants (12 TR [63%]; 13 DD [59%]) showed the typical adaptation response (decreasing F1 in response to an increased F1 during altered auditory feedback) whereas 16 (39%) participants (7 TR [37%]; 9 DD [41%]) did not. The total number of adapters and non-adapters did not significantly differ between groups (χ^2 = .071, p = .790). Figure 1 shows individual examples from the different phases of the experiment for a non-adapting individual with DD, a typical reading individual and an individual with DD. Independent t-tests showed that the adapters and non-adapters did not significantly differ on the reading and phonological tests (all p-values >.08). Also, the gender distribution within groups was not significantly different between adapters and non-adapters for the TR group (χ^2 = .022, p = .882), nor for the DD group (χ^2 = .087, p = .769).

Our second research question focused on the frequency of F1 productions across the different phases of the experiment for the adapters only (as shown in Figure 1), for which we performed linear mixed effects modeling. Group (TR vs DD), trial, gender (male vs female) and phase of feedback (baseline, ramp, hold, after-effect) were entered as fixed factors. As random effects, participants were added, along with by-participant slope adjustments for feedback phase and trial (Barr, Levy, Scheepers, & Tily, 2013). The best model fit was obtained by likelihood ratio tests using the maximum likelihood criterion. Satterthwaite approximations were used to estimate p-values within the model (Kuznetsova, Brockhoff, & Christensen, 2015). The resulting model had gender and a feedback by trial and a feedback by group interaction entered as fixed factors in the model. This model was significantly better than a model with gender and trial plus a feedback by group interaction ($p < .001$) and better than a model with gender, group and a feedback by trial interaction ($p < .001$). Adding three-way or four-way interactions did not significantly improve the model fit.

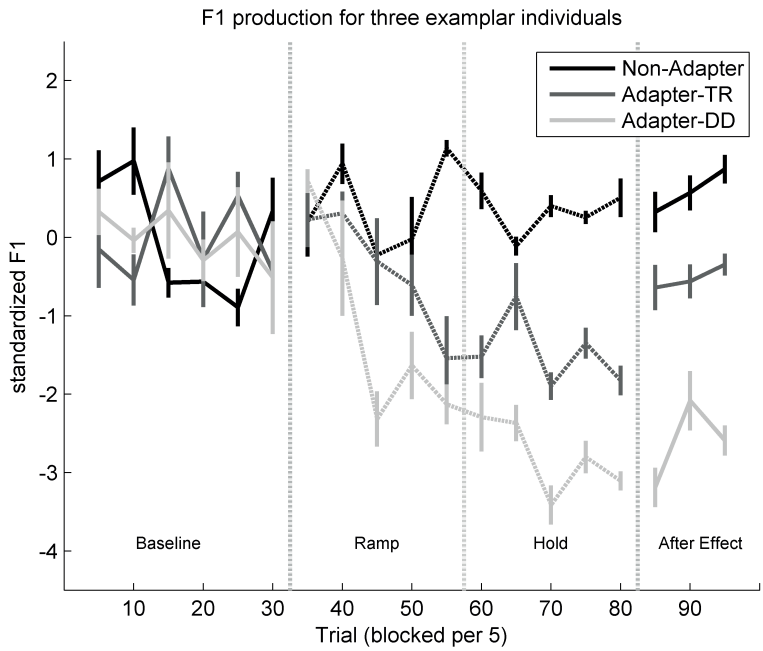

FIGURE 1. Individual examplars of a non-adapting individual, a typical reading individual and an individual with DD.

In accordance with the expectations, females had a significantly higher F1 production than men (β = 132.66, SE = 21.021, p < .001) and F1 production was significantly decreased in the hold phase (β = -22.30, SE = 6.011, p < .001) and after-effect phase (β = -24.49, SE = 6.052, p < .001). Trial had a small but significant effect on F1 production (β = -0.29, SE = .111, p < .01). No main effects were found for group, DD versus TR (β = -27.80, SE = 19.567, p = .168), and for the ramp phase (β = -1.54, SE = 4.007, p = .702). Significant feedback by trial interactions were found for the ramp phase (β = -0.66, SE = .159, p < .001) and the after-effect phase (β = 1.56, SE = .289, p < .001) but not for the hold phase (β = -.25, SE = .159, p =.123). Most interestingly, a stronger decrease in F1 frequency during the ramp phase (β = -9.67, SE = 4.63, p = .047), and a weaker increase of F1 frequency during the after-effect phase (β = -20.30, SE = 7.55, p = .013) was revealed for the DD group compared to TR group, while no significant group difference was observed during the hold phase (β = -9.86, SE = 7.754, p = .215). Including the non-adapters yielded an insignificant model, possibly due to the increased variance, but the general pattern remained the same for the after-effect. Importantly, the frequency of F1 productions during the baseline phase also did not differ between the TR-group (M_{F1} = 664.90 Hz; SD_{F1} = 70.08) and the DD-group (M_{F1} = 694.95; SD_{F1} = 76.96; t[23] = -1.018; p

= .319). Similarly, no significant difference in the variability of the productions between groups during the baseline was observed (TR: M_{SD_F1} = 20.23; SD_{SD_F1} = 8.33; DD: M_{SD_F1} = 20.32; SD_{SD_F1} = 6.67; t[23] = -.704; p = .489). All coefficients of the final model are summarized in table 2.

TABLE 2. The fixed effects coefficients of the final model (gender + feedback * trial + feedback * group)

Fixed effect	β	Std. Error	p
Intercept	*569.78*	*21.198*	*<.001*
Gender Females	*132.66*	*21.021*	*<.001*
Group DD	*27.80*	*19.567*	*.168*
Trial	*-0.29*	*0.111*	*<.01*
Feedback Ramp	*-1.54*	*4.007*	*.702*
Feedback Hold	*-22.30*	*6.011*	*<.001*
Feedback After-effect	*-24.49*	*6.052*	*<.001*
Feedback Ramp * Trial	*-0.66*	*0.159*	*<.001*
Feedback Hold * Trial	*-0.25*	*0.159*	*.123*
Feedback After-effect * Trial	*1.56*	*0.289*	*<.001*
Feedback Ramp * DD	*-9.67*	*4.629*	*.047*
Feedback Hold * DD	*-9.86*	*7.754*	*.215*
Feedback After-effect * Trial	*-20.30*	*7.55*	*.013*

To address our third research question, we performed a number of exploratory correlation analyses to investigate whether the slope and magnitude of the standardized response to the manipulation related to performance on reading and phonological awareness tasks. The slope of the response during the ramp and after-effect phase and the averaged response during the maximally altered feedback signal were correlated with the reading and phonological awareness scores for the sample as a whole, and for the DD and TR groups separately. The sample as a whole showed a significant correlation between the average frequency of the first formant during the hold phase and the number of reading errors made (r = -0.45, p = .022). A stronger response to the manipulation was related to more reading errors (see Figure 3a). The DD group showed significant correlations between the slope in the ramp phase and phonological awareness accuracy (r = 0.57, p = .044; Figure 3b), and between the slope in the after-effect phase and the number of reading errors (r = -0.64, p = .019; Figure 3c) and reading time (r = -0.71, p = .007; Figure 3d). A steeper slope away from baseline in

the ramp phase and a shallow slope towards baseline in the after-effect phase were thus associated with poorer phonological and reading abilities in the DD group. The control group did not show any significant correlations.

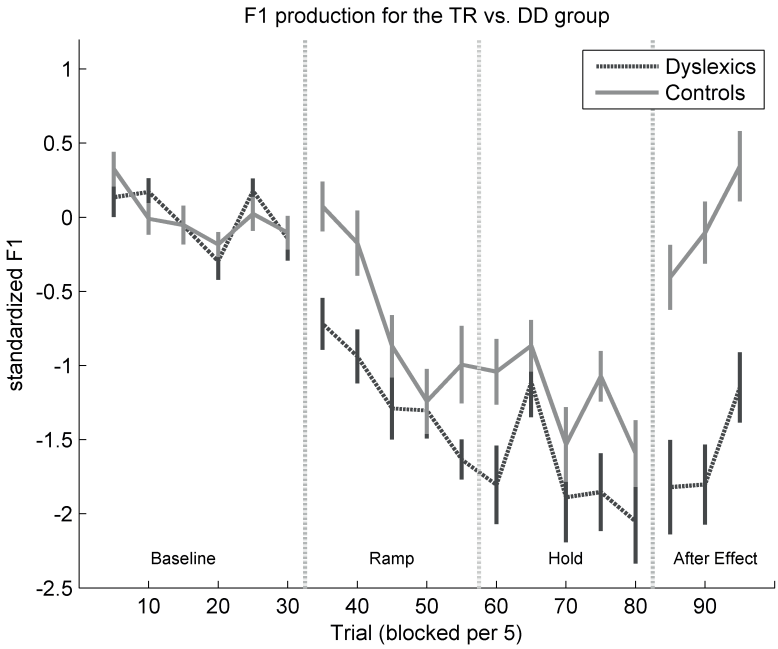

FIGURE 2. The adapted responses to the altered auditory feedback over the course of the experiment for adults with developmental dyslexia (DD; dashed line) and typically reading controls (TR; continuous line). Plotted is the average frequency of the first formant per group, averaged per 5 trials. Error bars represent ± 1 SE.

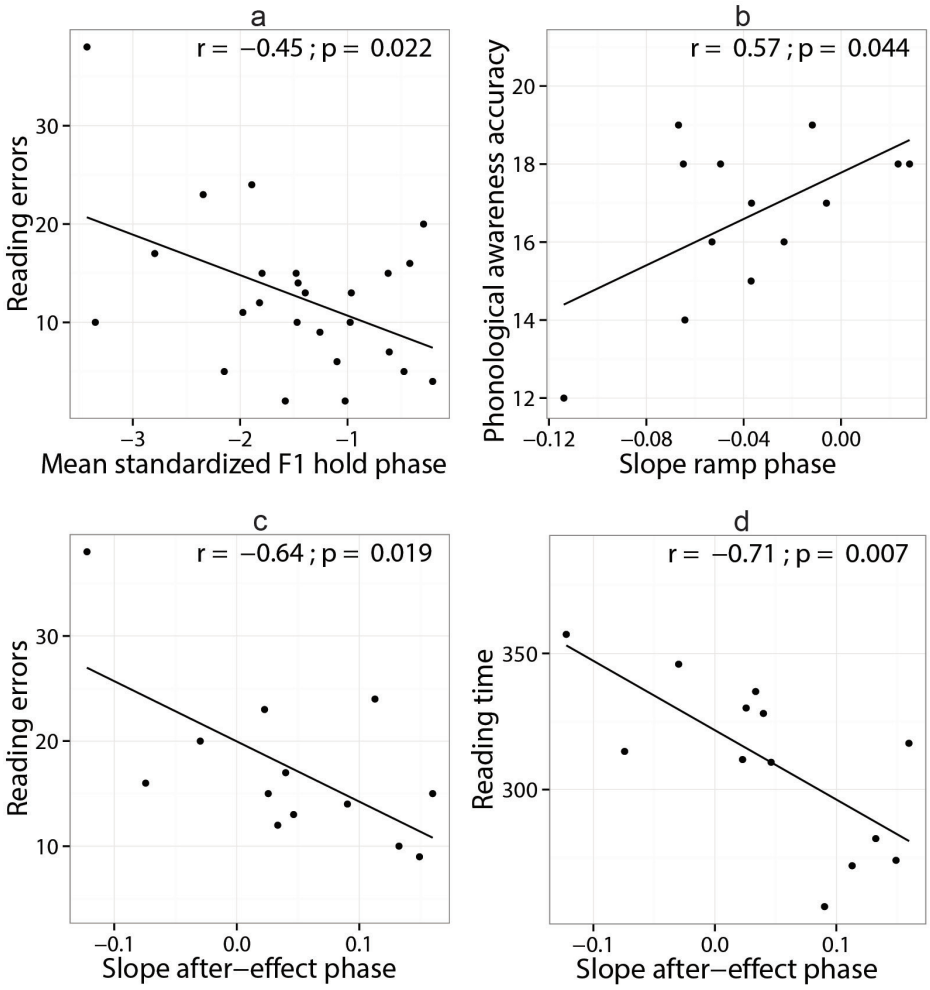

FIGURE 3. Correlations between (a) the averaged response during the hold phase and reading errors for the whole sample (n = 25); the slope in the ramp phase and phonological awareness accuracy (b), the slope in the after-effect phase with reading errors (c) and reading time (d) for the DD-group only (n = 13).

DISCUSSION

In this study, we examined speech feed-forward and feedback mechanisms—assumed to be critical for the formation and adjustment of phonological representations—in adults with developmental dyslexia and typically reading controls, using an altered auditory feedback paradigm. We found that about 61% of all participants showed the typical adaptation response to the F1 increase in the altered feedback phase. The number of adapters and non-adapters did not significantly differ between the two groups, and this could not be explained by differences in behavioral abilities or gender distribution. Furthermore, it was evidenced that people with DD adapted stronger in the ramp phase, and returned to baseline to a lesser extent when feedback was back to normal when compared to the TR-group. Finally, exploratory correlational analyses showed that a faster deviation from baseline during the ramp phase, stronger adapting response during the hold phase, and a slower return to baseline during the after-effect phase, were associated with poorer reading and phonological abilities.

This finding, that DD is related to stronger adaptation under conditions of altered feedback and to weaker de-adaptation when feedback is back to normal, is not fully explained by the allophonic theory of speech perception in DD discussed in the introduction. This view predicts that individuals with DD are (unconsciously) more sensitive to acoustic variation within a phoneme category. This explanation fits with the stronger adaptation in the ramp phase we did find, since the perturbation in this study remained within a phoneme category. Arguing against this explanation is the lack of a significant difference between groups for the hold phase. If individuals with DD have an allophonic mode of speech perception, a large difference should be present also for the hold phase. Even more importantly, we think this explanation predicts a stronger return to baseline for the people with DD, while the opposite was found. The stronger response to altered auditory feedback in the ramp phase is at least partially consistent with an overreliance on auditory feedback, another model discussed above. This interpretation corresponds to neurocomputational models showing that stuttering, reported to share characteristics with DD (Malek et al., 2013), is characterized by a bias towards feedback control (Civier et al., 2010). However, this overreliance on auditory feedback could also not explain the weak de-adaptation in DD in the after-effect phase.

Additionally, the current study does not support the claim that the phonological deficit should be reformulated as entirely a deficit in access to phonological representations, as proposed by Ramus and Szenkovits (2008) and Boets et al. (2013). First, Ramus and Szenkovits (2008) claimed that the phonological deficit becomes apparent when tasks place a strong demand on short-term memory, conscious awareness and speed. The

task employed in this study requires very minimal short-term memory, has no time constraints and the evoked response, when present, remained completely unconscious for all participants. Second, if there is an (unconscious) impaired access to phonological representations, individuals with DD should be less susceptible to alterations in feedback in this perception/production task, while the opposite is found for the ramp phase and no differences were found in the hold phase. This study shows in a novel way that the quality of the representations themselves is impaired in DD. Again, although deficits in access and retrieval are characteristics of DD as Ramus and colleagues suggest (2008), these findings do also suggest suboptimal phonological representations given the full set of findings.

As noted in the introduction, there are models that may be more parsimonious here. Thus, it may be that the current results fit best with the framework of the native language magnet theory (Kuhl, 1991; Feldman, Griffiths, & Morgan, 2009; Guenther & Gjaja, 1996), which claims that a phonetic category prototype functions as a perceptual magnet, resulting in poorer discriminability for neighboring stimuli close to the prototype. Importantly, a recent update of the theory (Native Language Magnet theory – expanded; Kuhl et al., 2008) argues for a strong interaction between the perceptually formed representations and their associated speech production traces. According to Kuhl and colleagues (2008) the development of motor commands is based on vocal imitation and experience with language. The results of articulatory movements are related to acquired auditory targets which yield a 'learned mapping' between the auditory and articulatory targets. Deficiencies in the perceptual warping of the acoustic space hampers this mapping and could hence affect the adequacy of both the feedback and feed-forward system. As such it might be more appropriate to conceptualize the magnet as a sensorimotor magnet, rather than purely a perceptual magnet. Recently, Niziolek and Guenther (2013) provided evidence that the response to altered auditory feedback is significantly influenced by the perceptual magnet effect, with responses up to three times bigger for near-phoneme boundary compared to near-phoneme center perturbation. The results of the current study suggest that the phonological deficit in DD is associated with a 'weaker' magnet (i.e. deficient warping), which makes it easier for individuals with DD to be moved away from the prototype (hence, stronger adaptation) and harder to return to baseline (hence, weaker de-adaptation). In addition, a weaker magnet in DD does not necessarily suggest a difference in the hold phase of the current experiment, which is in correspondence with the results of this study. Once the magnet loses its attracting influence on the perception and production of the utterance, all individuals plateaued at a similar amount of adaptation for both groups. An alternative account for the non-significant difference during the hold phase is that the phonological adaptation has not reached its maximum. Lametti et al. (2014) showed that individuals

continue to adapt for at least 200 trials. Interestingly, this weaker magnet theory not only offers an explanation for why individuals with DD perform more poorly on speech identification tasks (for instance during speech-in-noise tasks; Ziegler et al., 2009), but also why individuals with DD perform better at discriminating stimuli in conditions 'where phonemic categories are weakly perceptible' (Serniclaes et al., 2004). Forming stable grapheme-phoneme associations is a crucial step in reading development (Puolakanaho et al., 2007) and a weaker magnet could hinder the formation of these associations. More specifically, if a grapheme is coupled to a variant sensorimotor target (i.e. less strongly attracted to the center of a phoneme category) the grapheme-phoneme association will be noisier, and hence less efficient.

A number of steps should be undertaken in future research to further support this finding and to disentangle the contribution of the different explanations. Firstly, the current study applied altered auditory feedback only and approximately 39% of the participants did not show the typical adaptation response. Although this percentage of non-adapters is not abnormal (e.g. Lametti et al., 2012; Ito, Coppola, & Ostry, 2016)), a relatively high number of participants had to be excluded from the analyses. This might affect the generalizability of the findings to the DD population as a whole. Nonetheless, since non-adapters are equally distributed across groups, the non-adapting response seems not to be related to DD. The adaptation applied in this study was in the direction of a non-existing vowel in the Dutch language and we believe this could (partly) explain the percentage of non-responders. Including altered somatosensory feedback as a condition would likely allow analyzing the response for almost all participants (Lametti et al., 2012). Moreover, it will also indicate whether the current results are restricted to the auditory modality or extend to somatosensory feedback. The latter has been suggested for stuttering (Civier et al., 2010) but needs more thorough examination.

Secondly, it is important to note that in our study the perturbation of auditory feedback did not cross a phoneme boundary. The manipulation increased the first formant of the /ɛ/ vowel in the direction of the English /ae/ vowel, which is not existing in Dutch. Crossing phoneme boundaries not only increase the magnitude of the response (Niziolek & Guenther, 2013), but would also allow us to elucidate on both the weaker magnet hypothesis and the allophonic perception theory of DD (Serniclaes et al., 2004). Future studies could include both within and across phoneme boundary perturbations. A larger difference in adaptation between DD and TR groups for the within phoneme boundary manipulation as compared to the across boundary manipulation during a ramp phase, could be taken as corroborating evidence for an allophonic perception mode. The native language magnet hypothesis suggests that at a certain point after crossing the phoneme boundary, the altered percept should be attracted to the other

phoneme and this probably results in stronger compensation. A relatively stronger response for a perturbation crossing a phoneme boundary in the TR group as compared to the DD group could be taken as supporting evidence for the weaker magnet in DD. In addition, and in order to more explicitly examine the complementarity of or contradiction between these different characterizations of the phonological deficit, future studies should include measures that directly assess the allophonic mode of perception and of phonological access.

Thirdly, as expected, individual differences in phonological and reading abilities were associated with the response to altered auditory feedback. A stronger response was found to be associated with poorer phonological and reading skills. In this study, we did not include established and explicit measures of speech perception, speech production or non-word repetition. Relating these measures to the response to altered auditory feedback could further elucidate on speech perception-production interaction impairments in DD and could clarify the nature of the phonological deficit.

Lastly, individual differences in the direction and magnitude of the effect should also be examined across different developmental phases. The present results are obtained in an adult population. However, the speech perception-production interaction is thought to be particularly crucial in early development in which phonological representations are formed and established. Similar studies should be conducted in several childhood populations (e.g., typical, at-risk for DD, before and after literacy instruction, etc.) to see whether, when, and how speech feed-forward and feedback mechanisms are involved in the adequate development of phonological representations.

To our knowledge, this is the first study that explicitly investigates whether and how speech perception-production interaction is malfunctioning in DD. We reported that people with DD adapted more strongly in response to altered auditory feedback and de-adapted more weakly when feedback was back to normal, and that individual differences in this response were associated with phonological and reading abilities in adults with DD. We interpret these results as evidence for a weaker magnet in DD that is reflected in weaker sensorimotor attraction to the center of the phoneme category. Although it is clear that much work is needed to establish this finding in different populations with improved methodologies, this study opens a promising new line of research into the origin(s) of DD.

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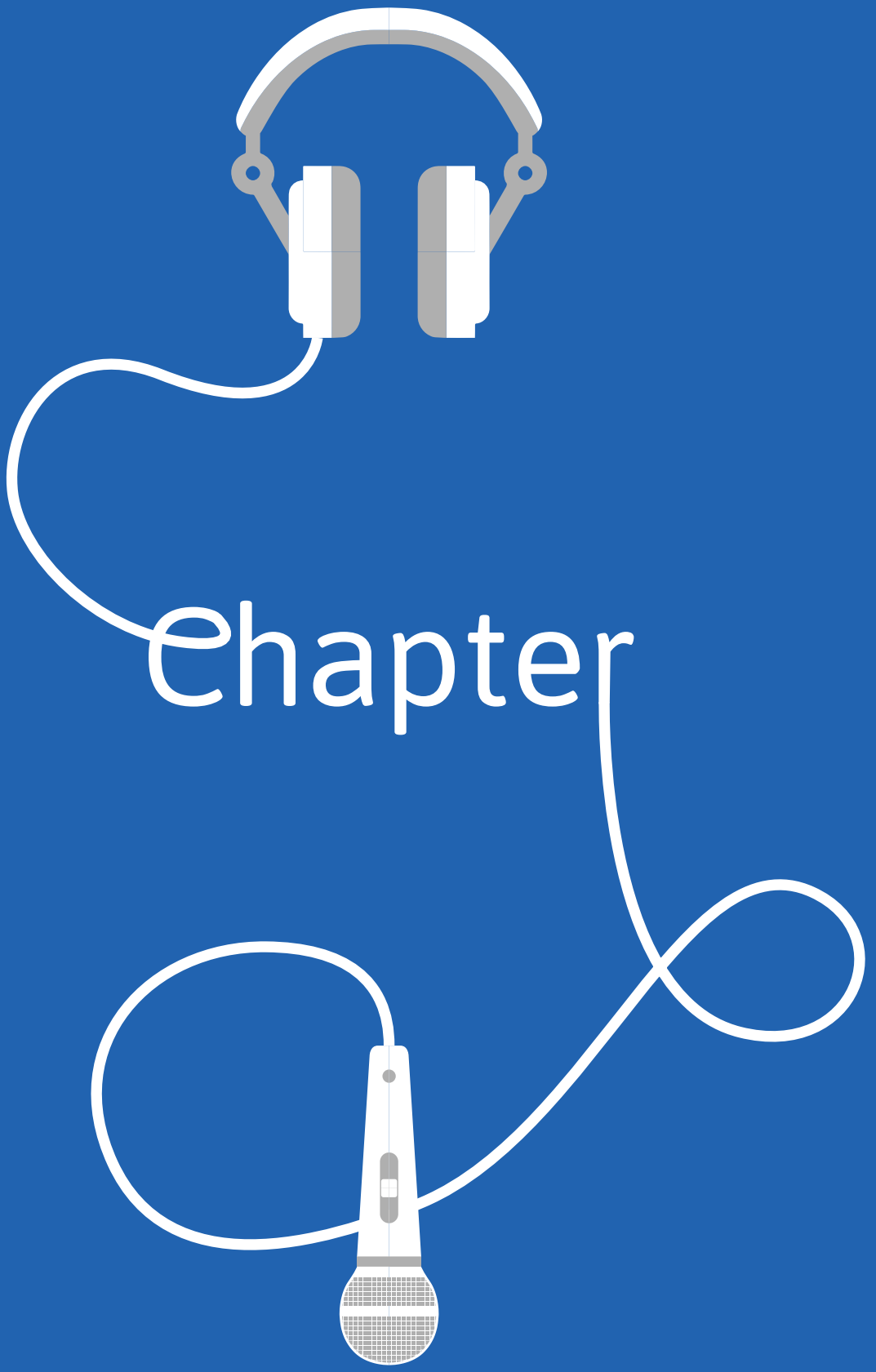
Chapter

NEUROBIOLOGICAL EVIDENCE FOR DEFICIENCIES IN THE SPEECH NETWORK IN DEVELOPMENTAL DYSLEXIA

Van den Bunt, M.R. [1*]
Francisco, A.A. [1]
Takashima, A. [2, 3]
Mars, R.B. [2,4]
Verhoeven, L [1]
Groen, M.A. [1]

[1] Radboud University, Behavioural Science Institute, Nijmegen, The Netherlands
[2] Radboud University, Donders Institute for Brain, Cognition and Behaviour, Nijmegen, The Netherlands
[3] Max Planck Institute for Psycholinguistics, Nijmegen, The Netherlands
[4] Centre for Functional MRI of the Brain (FMRIB), Nuffield Department of Clinical Neurosciences, John Radcliffe Hospital, University of Oxford, Oxford, United Kingdom
* Corresponding author: Mark van den Bunt; m.vandenbunt@pwo.ru.nl; phone nr.: +31243612430; Montessorilaan 3, 6525 HR Nijmegen, The Netherlands

3

ABSTRACT

It is hypothesized that Impaired phonological representations underlie dyslexia and that the development of adequate phonological representations depends on the interaction between speech perception and speech production mechanisms. In the current study, the neurobiological basis of these speech mechanisms was investigated. Twenty-one university students with dyslexia, and 19 typically reading students were recruited to participate in an MRI experiment. We examined (i) the fractional anisotropy of the arcuate fasciculus, (ii) the efficiency of the communication in a large speech network, and (iii) functional activation patterns for speech perception and production. We found developmental dyslexia to be characterized by deficiencies in the arcuate fasciculus, by less efficient communication between speech-related areas during resting-state scans and by a smaller extent of recruited speech sensory areas during speech production. This set of findings stresses the importance of taking the dynamics between speech perception and production into account, when examining phonological representations in dyslexia.

INTRODUCTION

Developmental dyslexia is a brain-based disorder in the acquisition of accurate and/or fluent word reading skill (Lyon, Shaywitz, & Shaywitz, 2003). Impaired quality of, or access to, phonological representations is often hypothesized to underlie this disorder (Elbro, 1998; Ramus & Szenkovits, 2008). Indeed, many studies have shown that both adults and children with dyslexia perform worse on tasks that rely on phonological awareness (Melby-Lervag, Lyster, & Hulme, 2012)—the ability to manipulate sounds in a spoken utterance. This meta-cognitive deficit in dyslexia is often attributed to deficiencies in speech perception and much effort has been put into defining this perceptual deficit (Noordenbos & Serniclaes, 2015; Ziegler, Pech-Georgel, George, & Lorenzi, 2009). Studies that examine speech production deficits in dyslexia showed that oromotor (Malek, Amiri, Hekmati, Pirzadeh, & Gholizadeh, 2013) and articulatory (Catts, 1986, 1989; Smith, Roberts, Lambrecht-Smith, Locke, & Bennett, 2006) skills may also be impaired in dyslexia. It is important to note that neurocomputational models of speech production have indicated that the quality of phonological representations hinges on both speech perception and production, and more specifically, on the interaction between speech feedforward and feedback mechanisms (Guenther, Ghosh, & Tourville, 2006; Hickok, Houde, & Rong, 2011; Tourville & Guenther, 2011). In a recent study, Van den Bunt and colleagues (2017) showed that individuals with dyslexia indeed responded differently in conditions in which the interaction between speech perception and production is crucial. In that study, two groups of adults, one with dyslexia and another with typical reading abilities, produced speech under conditions of unaltered and altered auditory feedback. Adults with dyslexia responded more strongly to altered feedback and afterwards returned to their own baseline production to a weaker extent than typically reading adults. This study suggested that behaviourally, individuals with dyslexia show impairments in the speech perception-produciton interaction. In the present study, we aim to shed more light on the neurobiological underpinnings of the role of speech perception and production interactions in the quality of phonological representations in individuals with dyslexia. Therefore, we investigated: (1) structural connectivity, by means of the organization of the arcuate fasciculus between the classical speech areas of perception (i.e. Wernicke's) and production (i.e., Broca's); (2) measures of neural network integration of a well-specified speech network in terms of feedforward and feedback mechanisms; (3) functional activation patterns during the perception and production of speech within this network.

The arcuate fasciculus

Two areas that have traditionally been seen as important for perception and production, are Wernicke's and Broca's areas, respectively. A classical model of the neurobiology of language is the Wernicke-Lichtheim-Geschwind model. This model associates Wernicke's area, located in the posterior superior temporal gyrus, with spoken word recognition (Penfield & Roberts, 1959) and Broca's area (inferior temporal gyrus) with speech production (Geschwind, 1965; see Levelt, 2013 for an overview of these models). However, this account is not considered tenable anymore (Hagoort, 2014). Instead, damage to the left STG is also associated with impaired speech production (Damasio & Damasio, 1980) and Broca's area has been implied in, amongst others, language comprehension (Grodzinsky & Santi, 2008). An influential model of the cortical organization of speech processing proposes a central role in sensorimotor control of speech for the dorsal stream (Hickok & Poeppel, 2007). The arcuate fasciculus (AF) is a white-matter bundle that structurally connects these superior temporal and inferior frontal areas (Catani & de Schotten, 2008) and this tract is likely to be important for this dorsal stream. A direct pathway between Wernicke's and Broca's area and two indirect (anterior and posterior) segments is usually distinguished within the arcuate fasciculus (Catani, Jones, & Ffytche, 2005). Importantly, an electrocorticography study showed bidirectional communication along the arcuate fasciculus, providing a functional indication that this tract could serve a network of both feedforward and feedback projections in the perception and production of language (Matsumoto et al., 2004).

The arcuate fasciculus has been examined in many speech related studies, but notably, has also been a prime neuroanatomical target for research on the neurobiological basis of dyslexia (e.g. Feldman, Yeatman, Lee, Barde, & Gaman-Bean, 2010; Lebel et al., 2013a). In the context of reading research, the brain areas connected by the arcuate fasciculus are related to grapheme-to-phoneme mapping and phonological processing (left posterior superior temporal gyrus, left supramarginal gyrus) and articulation and naming (left inferior frontal gyrus). Several studies have compared the fractional anisotropy—a measure of diffusion that reflects several anatomical features of the organization of white matter (Beaulieu, 2009)—of the arcuate fasciculus in typical and dyslexic readers, but results are mixed. Some studies report that impaired reading abilities are related to reduced fractional anisotropy in the left arcuate fasciculus (Feldman, Yeatman, Lee, Barde, & Gaman-Bean, 2010; Gullick & Booth, 2015; Langer et al., 2015; Lebel & Beaulieu, 2009; Qiu, Tan, Zhou, & Khong, 2008; Rimrodt, Peterson, Denckla, Kaufmann, & Cutting, 2010; Vandermosten, Boets, Poelmans, Sunaert, Wouters, & Ghesquire, 2012), however, others failed to find this relation (Andrews et al., 2010; Dougherty et al., 2007; Rollins et al., 2009), or reported bilateral reductions in the fractional anisotropy of the arcuate

fasciculus in people with reading deficits (Lebel et al., 2013b; Steinbrink et al., 2008). Since the communication between these temporoparietal and frontal areas is both implicated in reading and in speech perception-production interactions, the current study will examine whether dyslexia is characterized by reduced structural (i.e. fractional anisotropy) connectivity of the arcuate fasciculus.

A more recent speech model

Although the Wernicke-Lichtheim-Geschwind model has been highly influential in thinking about the neurobiological basis of speech, more recent computational models on speech production greatly increased the precision of brain regions involved, and their interactions. Specifically, the Directions Into Velocities of Articulators (DIVA) model (Guenther et al., 2006; Tourville & Guenther, 2011) is an established and biologically plausible model on how speech feed-forward and feedback mechanisms relate to phonological representations and is well-specified in terms of hypothesized brain areas underlying these relations. The major components of this model include maps of phonological representations (in the left posterior inferior frontal gyrus or ventral premotor cortex), auditory target and error maps (Heschl's gyrus, posterior superior temporal gyrus), somatosensory target and error maps (ventral primary sensory cortex, supramarginal gyrus), and motor/articulatory maps (ventral primary motor cortex). Examining the functional integration—defined as the ability of the brain to efficiently and rapidly process and combine information of distributed brain regions (Rubinov & Sporns, 2010)—of these brain areas, both during resting-state, as well as during speech perception and speech production will further elucidate the neural basis of the phonological deficit in dyslexia. Graph-theoretical analyses are often employed to measure the efficiency of neural communication between different brain regions (Finn et al., 2014; Fraga González et al., 2016). These analyses include measures of functional integration, which reflects the ability to effectively combine information from distributed brain regions, such as the mean shortest path length between all nodes in the network or the global efficiency of the network (for an overview of network measures see Rubinov & Sporns, 2010). A recent EEG-study on whole-brain connectivity differences between typical readers and individuals with dyslexia showed, amongst other findings, increased connectivity from and towards central (i.e. pre- and postcentral) brain areas during word reading, and this was hypothesized to reflect an overreliance on temporo-parietal regions involved in sensorimotor control (Žarić et al., 2017). In order to examine the neural dynamics that underlie speech perception-production interactions, we examine in the current study whether dyslexia is characterized by differences on measures of functional integration in the neural correlates of the DIVA network.

Functional activation in the speech network during perception and production

Lastly, impairments in the interaction between speech perception and production could be more visible in the activation patterns during the perception or production of speech rather than during the resting-state. The majority of the brain activation studies studying the phonological deficit that have been done in dyslexia report that the deficit is related to brain areas involved in phonology (Dole, Hoen, & Meunier, 2012; Hernandez et al., 2013) or orthography to phonology conversion (Paulesu, Danelli, & Berlingeri, 2014). Particularly the occipito-temporal regions—associated with whole-word recognition and grapheme-phoneme couplings—are strongly recruited during reading in typical readers and hypoactivated in readers with dyslexia (Diehl et al., 2014; Pugh et al., 2000). More recent studies demonstrated, in typical readers, that reading ability could be explained by the extent to which reading and speech circuits overlap (Preston et al., 2015; Rueckl et al., 2015). However, most functional neuroimaging studies on phonological processing in dyslexia are informed by perceptual processing and/or the manipulation of phonological information, while reading is strongly related to overt production—at least during acquisition. A small number of studies included the production of speech in the fMRI task. For instance, Eden et al. (2004) compared participants with dyslexia and with typical reading abilities to repeat words. This condition, however, was only used as a contrast for repeating words with the removal of the first letter, a phonological manipulation task. A PET-study by McCrory, Frith, Brunswick, and Price (2000) examined the neural activation for repeating words and found that typical readers, compared to individuals with dyslexia, showed increased activation in cerebellar, superior temporal, and primary somatosensory regions. However, the repetition of words was contrasted with a rest condition, which meant they could not assess whether the differential activation patterns are caused by differences in speech input processing, output processing, or a combination of both. In this study the functional activation patterns during the perception and production of simple consonant-vowel syllables will be compared between groups. Particularly of importance is whether and how neural activation differences for these conditions are localized in speech sensory and production brain areas.

The present study

In short, studying the neurobiological underpinnings of perceptual processes alone may be too limited to further illuminate the phonological deficit in dyslexia. Therefore, in the current study we focus on structural and functional connectivity within a speech network that incorporates perception, as well as production and their interaction in adults with dyslexia relative to typical readers. First, we examined whether the

communication between Wernicke's and Broca's areas is impaired in readers with dyslexia, using structural connectivity measures of the arcuate fasciculus. Second, differences in network quality measures of functional integration, using the brain locations of an explicit model of speech production, were examined in a resting-state condition, as well as during speech perception and production. Lastly, we studied whether dyslexia was characterized by differences in brain activation during the perception and production of speech. We hypothesize that dyslexia is characterized by a deficit in white matter organization of the arcuate fasciculus, less effective communication in a functional speech network, and decreased functional activation in speech and language related areas during speech perception and production.

METHODS

Participants

Twenty-one university students with dyslexia (16 women; M_{age} = 24.20 years; SD_{age} = 2.57 years) and nineteen university students with typical reading abilities (12 women; M_{age} = 25.00 years; SD_{age} = 4.03 years) were recruited to participate in the study. Groups did not differ in terms of age (t[30.05] = 0.737, p = .467) or in gender distribution (χ^2 = .306, p = .580). All participants were native Dutch speakers and participated in an earlier behavioral study in which reading abilities were examined. Inclusion criteria for the dyslexia group were an official diagnosis and reading performance below the 30th percentile on reading accuracy or reading time on a standardized reading test (GL&Schr – *Test voor gevorderd Lezen en Schrijven* [Test for Advanced Reading and Writing; Depessemier & Andries, 2009]). No diagnosis of dyslexia and no history of reading difficulties, in addition to a score above these thresholds was required to be included in the group with typical reading abilities. Individuals with typical reading abilities outperformed the dyslexia group in terms of reading accuracy (t[19.94] = 5.04, p <.001) and reading speed (t[21.37] = 7.83, p <.001). In addition, all participants passed an audiometric test in which pure tones should be detected at less than 30 dB at 250 Hz, 500 Hz, 1 kHz, 2 kHz, and 4 kHz in both ears. Three participants (2 with dyslexia; 1 typical reader) were excluded for the DTI analysis due to corrupted data (excessive motion, poor tensor fitting). Participants provided informed consent prior to participation and received a monetary compensation afterwards. This study was conducted with approval of the ethical committee for human medical research (CMO Arnhem-Nijmegen, the Netherlands).

Data acquisition

All images were acquired using a 3T MAGNETOM Trio PRISMA^{fit} system (Siemens Healthcare, Erlangen, Germany) with a 32-channel head coil. Noise-cancelling earphones were provided to reduce the influence of scanner noise and to present auditory stimuli, and foam pads were used to limit head motion. A high-resolution T1-weighted 3D ultrafast echo scan was acquired using the MPRAGE sequence (176 slices; slice thickness = 1.0 mm; TR =2400 ms; TE = 2.13 ms; FA = 8°; FOV = 256 x 256 mm; voxel size = 1 x 1 x 1 mm^3). A field-map scan was acquired to correct for inhomogeneity in the magnetic field. A 2-shell NODDI protocol (Zhang, Schneider, Wheeler-Kingshott, & Alexander, 2012) was used to obtain diffusion-weighted images (10 unweighted images; a 30-direction shell at b = 750 and a 60-direction shell at b = 3000; TE = 7300 ms; TR = 77 ms; matrix size = 990 x 990; voxel size = 2.00 x 1.98 x 1.98 mm^3). An 8-multiband accelerated whole-brain fMRI acquisition was used to acquire data for functional activity and resting-state connectivity (TR = 735 ms; TE = 39 ms; FA 52°; voxel size = 2.4 x 2.4 x 2.4 mm^3; 1200 timepoints for the functional task; 700 for the resting-state data).

Materials and Procedure

Stimuli

The same thirty phonotactically legal but meaningless (both in Dutch and in English) consonant-vowel stimuli were used in the speech perception and production tasks. For speech perception, these stimuli were recorded in a sound attenuated room at 44.1 kHz with a Sony Handycam DCR-SR190E, produced by a female adult native speaker of Dutch and were presented to the participants via MR-compatible earphones. The stimuli for the speech production task were presented orthographically with white letters on a black background. The stimulus materials were delivered using Presentation software (Neurobehavioral Systems; www.neurobs.com). Each stimulus was presented once per condition. A list of the used stimuli is provided in Appendix 1.

Tasks during fMRI

A block-design one-back task was employed in the scanner and this task consisted of six conditions (words, pseudowords, illegal letter strings, audiovisual speech perception, auditory-only speech perception, and speech production) of which only the results of the auditory-only speech perception condition and the speech production condition are reported here (the other conditions will be reported in a separate paper on audiovisual integration in dyslexia). Blocks of stimuli were presented five times for each condition in a pseudorandomized order so that no condition appeared twice in a row. Each speech perception block consisted of seven stimuli (of which one was a repetition) with a duration of 1500 ms each, and a 500 ms inter-stimulus interval. Each

speech perception block thus lasted approximately 14 s. Each speech production block consisted of six stimuli with a duration of 2000 ms and a 500 ms inter-stimulus interval. Each block started with the presentation of an icon for 2500 ms, which indicated whether the following stimuli would be speech perception or speech production stimuli. The stimulus length for the speech production condition was slightly longer to allow for at least two and maximally three productions of the presented syllable. Each speech production block lasted for approximately 15 s. The 30 stimuli were shuffled for each condition and randomly distributed across the separate blocks. The inter-block interval was used as baseline and consisted of a fixation cross on the screen for 16 s. To ensure participants attended to the stimuli, participants were instructed to press a button whenever the same stimulus was presented twice in a row, which occurred once in each block at a random position. The speech production task was thus effectively a dual task situation (i.e. production speech and pressing a button). Since we wanted to ensure that differences between conditions in (speech) motor areas were not related to a button press, we included the button press in all conditions. The volume of the auditory stimuli was adjusted for each participant prior to the task such that the stimuli were clearly audible above the scanner noise.

During the resting state scan, participants were instructed to fixate on a white fixation cross on a black background, using a resolution of 1024 x 768 and font size of 48 points, and not to think of anything in particular. This procedure is reported to yield the most reliable data (Birn et al., 2013; Patriat et al., 2013).

Participants were instructed to lie as still as possible during all scans (structural and functional). The scanning session took approximately 45 minutes.

Preprocessing and analysis

The diffusion weighted images (DWI) were preprocessed using the standard pipeline of the FMRIB Diffusion Toolbox in FSL (http://www.fmrib.ox.ac.uk/fsl). In short, raw dicom images were first converted to a 4D nifti file. The data was subsequently corrected for eddy currents and skull stripped. A crossing fiber model was fitted to the data using a multi-b-value model using Bayesian Estimation of Diffusion Parameters Obtained using Sampling Techniques (*Bedpostx*) which runs Monte Carlo sampling to obtain distribution on diffusion parameters at each voxel. The interpretation of fractional anisotropy (FA) of the traditional diffusion tensor model can be ambiguous in regions where fibres are crossing. The tbss_x procedure was run to ensure that the anisotropy measures were all of fibres in the same orientation (Jbabdi, Behrens, & Smith, 2010). The fractional anisotropy measures of the first fiber were registered to the 1 x 1 x 1 mm^3 standard space included in the FSL toolbox using a nonlinear registration. Next,

the images with the mean fractional anisotropy of the principal fiber (referred to as FA1) were masked for the arcuate fasciculus using the template of Catani et al. (2005) and thresholded at the default value of .2. A voxel-wise statistical analysis was applied on the FA1 data using Tract-Based Spatial Statistics (Smith et al., 2004, 2006). A mean FA1 image was created and thinned to represent the center of the tracts and masked by a binarized mask of the arcuate fasciculus (Catani et al., 2005) to restrict the analysis to our tract of interest. The FA1 data of each subject was subsequently aligned to this mean image and the resulting data was fed into the between-subjects voxel-wise statistics. A voxel-wise comparison with $p < .005$, and a cluster of more than ten voxels, was used to reveal differences between typical readers and individuals with dyslexia.

Preprocessing of the functional data (task and resting-state) was done using SPM8 (Statistical Parametric Mapping; Wellcome Department of Cognitive Neurology, London, UK; www.fil.ion.ucl.ac.uk). The data were realigned and unwarped, field-map corrected, segmented and normalized to 2 x 2 x 2 mm^3 MNI space. The effects of head motion were regressed out for all functional data using 6-motion parameters. A 3D Gaussian kernel (8 mm full width at half maximum) was used to spatially smooth the functional images. The resting-state and task-based connectivity data was bandpass filtered (0.008 – 0.09 Hz). In agreement with the protocols of the Human Connectome project (humanconnectomeproject.org), multiband data were not slice-time corrected.

Next, for the resting-state fMRI functional connectivity analyses, the hemodynamic response in 62 regions of interest (ROI), representing the neural substrates of all components of the DIVA model (Guenther et al., 2006; Tourville & Guenther, 2011, using the presented MNI coordinates with a 5 mm surrounding sphere), were compared between groups. A few ROIs (see Appendix 2) were surrounded with a smaller (3 mm) sphere, to avoid overlapping regions. The Conn toolbox (Whitfield-Gabrieli, & Nieto-Castanon, 2012; http://www.nitrc.org/projects/conn) was used to obtain Z-transformed correlation values between these 62 ROIs during the resting state scans and during perceiving and producing speech. These data were imported in the GAT toolbox (Hosseini, Hoeft, & Kesler, 2012) in which individual functional network measures for each group were calculated. Since there is no consensus on which density is best for comparing networks between groups, a range of densities was included in our analysis (densities between .15-.45 with .01 increments). Group differences between measures of functional integration (i.e. global efficiency, path length) were examined using an area under the curve comparison for the included densities between groups.

The task-related fMRI data were analyzed using a general linear model and statistical parametric mapping. Brain activation patterns between groups were compared for speech perception and speech production contrasted with baseline (fixation cross). A whole-brain analysis (cluster-corrected threshold for family wise error rates at $p < .05$, with a cluster determining voxel level threshold of $p < .001$, uncorrected?) was conducted to determine overall group differences in brain activation during speech perception and production. Xjview (http://www.alivelearn.net/xjview) was used to visualize functional activation patterns.

RESULTS

The arcuate fasciculus

A group comparison between the fractional anisotropy values on the skeletonized arcuate fasciculus revealed no differences for mean fractional anisotropy of the arcuate fasciculus as a whole (Welch's T-test: t[25.48] = 0.85, p = .402) or the left (Welch's T-test: t[28.04] = 0.75, p = .458) or right (Welch's T-test: t[23.81] = 0.90, p = .379) arcuate fasciculus, separately. Using a more sensitive approach, cluster-wise statistics revealed a higher local anisotropy in typical readers in two clusters in the arcuate fasciculus, a cluster of 25 voxels in the left and a cluster of 19 voxels in the right arcuate fasciculus. Figure 1 shows the thickened (for visualization purposes using tbss_fill) significant clusters on the mean FA1 image of the arcuate fasciculus (left panel) and the post-hoc comparison (right panel) of the left (Welch's T-test: t[33.87] = 5.88, p < .001) and the right (Welch's T-test: t[33.997] = 4.38, p < .001) cluster. Further exploration of the clusters, using the atlas of Catani and Mesulam (2008), showed that the significant differences were found on the long segment of the left arcuate fasciculus and the posterior segment of the right arcuate fasciculus, while no significant clusters were found for the left anterior and posterior segments and right long and anterior segments.

Neural network integration in a speech network

In these connectivity analyses we examined the functional integration of the speech network, consisting of 62 ROIs from the DIVA model, in individuals with dyslexia and with typical reading abilities. First, a 62 x 62 association matrix was generated for all subjects using the data from the resting state scans, to show the connectivity between the included ROIs (see Figure 2). Consistent with expectations, we observed that activity patterns of areas involved in speech production and perception were positively correlated, even in the case of spatially separated regions such as in the cerebellum and frontal cortex.

The functional integration measures *global efficiency* and *path length* did not show a significant difference between individuals with dyslexia and with typical reading abilities during speech perception (global efficiency: $p = .356$; path length: $p = .235$) and production conditions (global efficiency: $p = .089$; path length: $p = .436$). However, during the resting-state condition, individuals with typical reading abilities showed higher global efficiency ($p = .047$), and a lower path length lambda ($p = .009$) when compared to individuals with dyslexia. Figure 3 shows the difference in global efficiency and the normalized path length across the full range of densities.

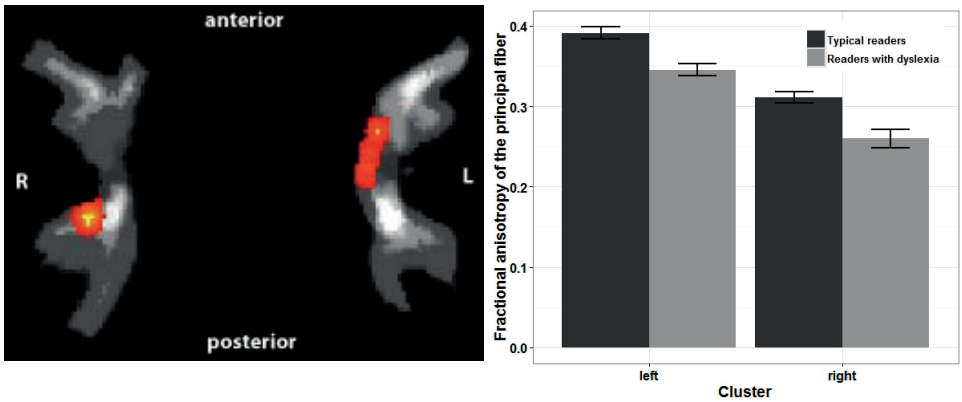

Figure 1. Group differences in fractional anisotropy of the arcuate fasciculus. Left panel: left and right arcuate fasciculus, with the tract intensity differences (gray-white) reflecting the fractional anisotropy (brighter is more anisotropy). On top of that the red-yellow intensity differences depict the significance of the group difference for that voxel (the more yellow the lower the p-value). Right panel: mean fractional anisotropy of the clusters in the left and right arcuate fasciculus for the readers with dyslexia (grey) and the typical readers (black). Error bars represent standard errors.

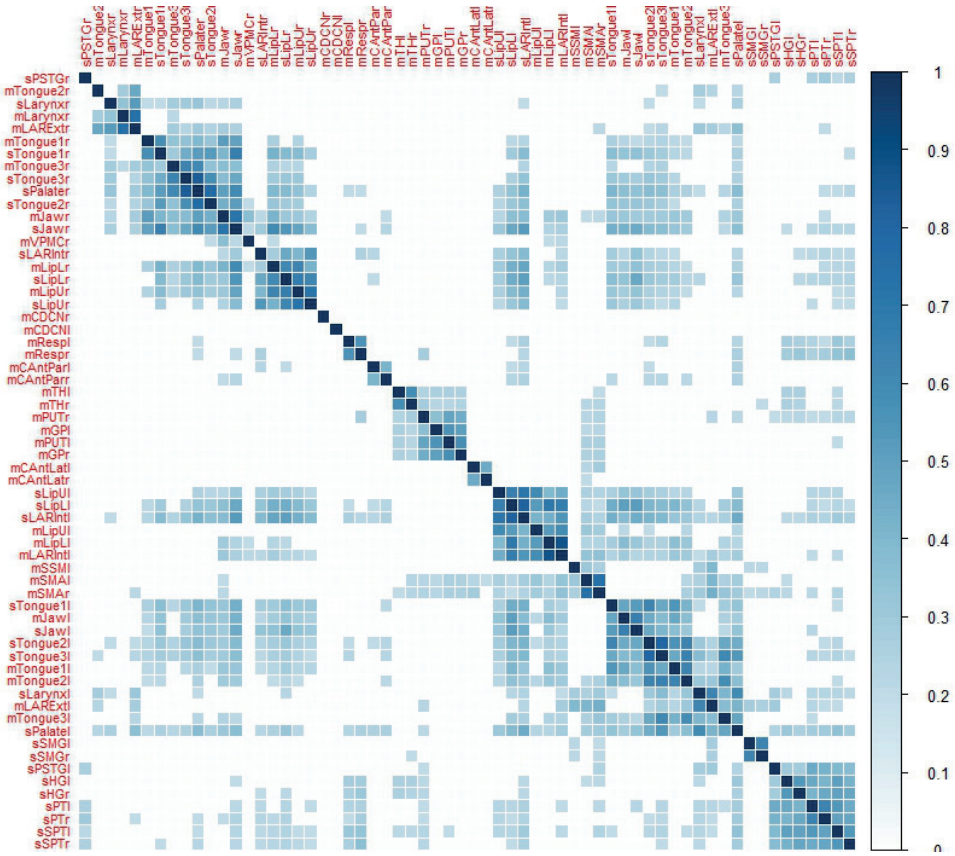

Figure 2. Association matrix for all participants during resting-state. All ROIs are derived from the DIVA model. The color bar represents a Pearson correlation coefficient. All ROIs that start with an /s/ or and /m/ are primarily implicated in sensory and motor processing of speech, respectively. A small letter /l/ indicates a left-hemispheric region, a small /r/ a right-hemispheric region. PSTG = posterior superior temporal gyrus; LarExt = larynx extrinsic; VPMC = ventral premotor cortex; LarInt = Laryinx Intrinsic; LipL = lower lip; LipU = upper lip; CDCN = Cerebellum Deep Cerebellar Nuclei; Resp = Respiration; CAntPar = Cerebellar Anterior Paravermis; TH = thalamus; PUT = putamen; GP = globus pallidus; SSM = speech sound map; SMA = supplementary motor area; SMG = supramarginal gyrus; HG = Heschl's gyrus; SPT = Sylvian parietal temporal.

Functional activation during speech perception and speech production

For the functional activation patterns during speech perception and production, we first tested whether possible differences were not due to differences in task performance. Both groups performed similarly on the speech perception ($M_{Dyslexia}$= 3.86; $SD_{Dyslexia}$ = 1.68; $M_{Typical}$ = 4.11; $SD_{Typical}$=1.10; Welch's T-test: t[34.76] = .557, p = .581)) and speech production ($M_{Dyslexia}$= 3.43; $SD_{Dyslexia}$ = 1.83; $M_{Typical}$ = 4.32; $SD_{Typical}$=1.33; Welch's T-test: t[36.43] = 1.761, p = .087) tasks. Next, contrast images were generated for speech perception over baseline and for speech production over baseline for each individual. These images were subsequently entered into second-level analyses using independent sample t-tests to compare the two groups. No significant clusters were found for the speech perception condition between groups. The speech production comparison between groups yielded five significant clusters, in the right hemisphere centered around auditory (superior temporal gyrus [STG]) and somatosensory areas (Primary somatosensory area[S1]), and in the left hemisphere around auditory (STG), somatosensory (S1, supramarginal gyrus [SMG]) and motor areas (supplementary motor area [SMA], rolandic operculum [RO]) that were greater for typical readers than for individuals with dyslexia. Table 1 and Figure 4 and 5 provide the characteristics of these significant clusters. The reverse contrast revealed no signifincant clusters. Next, we examined whether including measures of structural (fractional anisotropy in the significant cluster) and functional (measures of global efficiency and path length) connectivity accounted for this difference in activation. Including these measures yielded the same significant clusters with only marginal changes in cluster sizes.

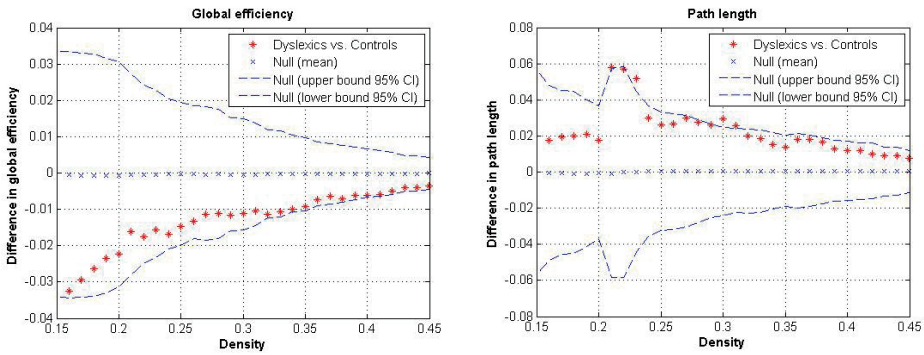

Figure 3. These plots depict the significant speech network differences for functional integration for global efficiency (left panel) and path length (right panel) between typical readers and individuals with dyslexia. Both results are based on resting-state data.

Table 1. Areas that showed reduced activation during speech production for individuals with dyslexia compared to typical reading abilities

Cluster-level			Peak-level				MNI	Area	Proposed role in speech*
p_{FWE}	q_{FWE}	k_e	p_{FWE}	q_{FWE}	T	Z			
<.001	<.001	629	.002	.030	6.87	5.53	64, -18, 0	rSTG	Auditory perception
			.285	.432	5.07	4.42	50, -2, -2	rSTG	Auditory perception
			.405	.432	4.90	4.30	50, -12, 4	rSTG	Auditory perception
<.001	<.001	803	.056	.259	5.73	4.85	-58, -12, 26	lS1	Somatosensory perception
			.162	.432	5.31	4.58	-62, -16, 12	lSTG	Auditory perception
			.268	.432	5.10	4.44	-52, -22, 36	lSMG	Somatosensory perception
<.004	.005	147	.409	.432	4.90	4.30	-4, -12, 68	lSMA	Initiating/sequencing speech
			.502	.432	4.79	4.22	-10, -6, 62	lSMA	Initiating/sequencing speech
			.961	.698	4.16	3.75	-16, 0, 66	lSMA	Initiating/sequencing speech
<.001	.001	193	.600	.505	4.68	4.14	-52, 2, 10	lRO	Not specified
			.823	.605	4.42	3.95	-40, -4, 12	lRO	Not specified
			.918	.637	4.27	3.84	-56, -2, 2	lSTG	Auditory perception
<.001	.001	216	.735	.551	4.53	4.03	58, -34, 42	rSMG	Somatosensory perception
			.894	.633	4.31	3.88	44, -40, 62	rS1	Somatosensory perception
			.964	.698	4.15	3.75	46, -32, 60	rS1	Somatosensory perception

Note: Table shows three local maxima more than 8mm apart per cluster. * = based on Guenther et al., 2006; Abbreviations: STG = Superior Temporal Gyrus; S1 = Primary somatosensory cortex; SMG = Supramarginal Gyrus; SMA = Supplemantary Motor Area; RO = Rolandic Operculum; lowercase r and l indicate left and right hemisphere

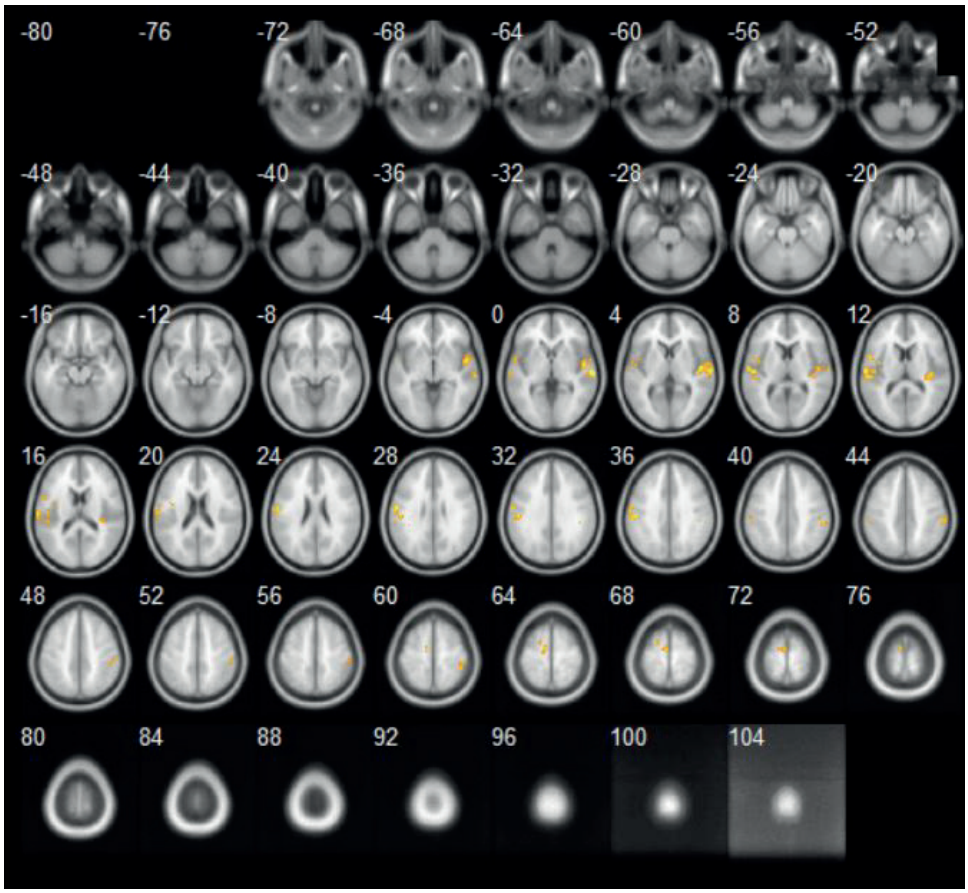

Figure 4. Slice view of the significantly stronger activation in typical readers compared to individuals with dyslexia during speech production. Figure is presented in neurological convention (left of the brain on right side of figure).

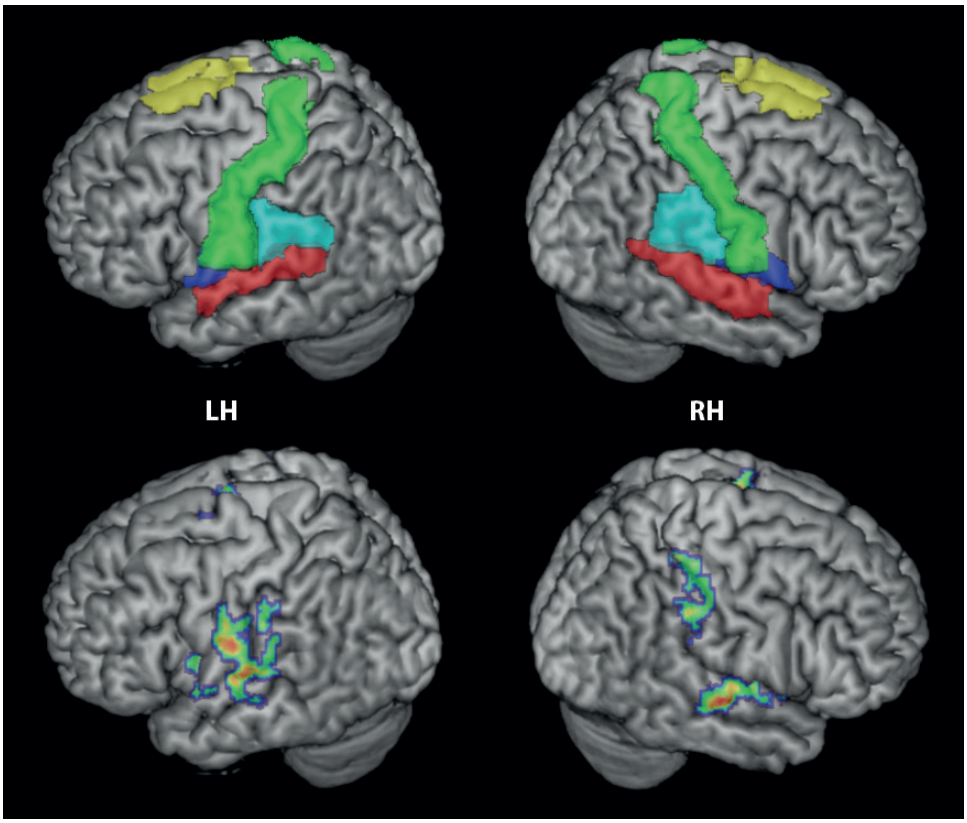

Figure 5. Lateral view of the clusters with significantly stronger activation for typical readers compared to individuals with dyslexia (bottom panels). Anatomical regions involved in speech processing in which peak activations of clusters are reported are highlighted in the top panels. Red = Superior temporal gyrus; Blue = Rolandic operculum; Cyan = Supramarginal gyrus; Green = Primary somatosensory area; Yellow = Supplementary motor area. LH = left hemisphere; RH = right hemisphere.

DISCUSSION

The current study examined the neurobiological basis of deficits in the speech network in dyslexia, using three different approaches to examine structural and functional connectivity. First, we found that individuals with dyslexia showed weaker structural connectivity between Wernicke's and Broca's area, in two clusters in the arcuate fasciculus, than individuals with typical reading abilities. Second, the connectivity analyses of a large and distributed speech network showed that dyslexia is characterized by less efficient and integrated communication during resting-state conditions, but not during speech perception and production. Third, functional activation patterns during speech perception showed no group differences between individuals with dyslexia and with typical reading abilities. Interestingly, individuals with typical reading abilities showed stronger activation of speech sensory areas when producing speech than individuals with dyslexia. These functional activation differences could not be accounted for by differences in structural connectivity or functional integration of the speech network. This set of findings sheds new light on a possible neurobiological basis of the phonological deficit in dyslexia and stresses the importance of taking speech production and the interaction between speech perception and production into account.

The structural connectivity analyses showed that dyslexia is characterized by a reduced fractional anisotropy of the arcuate fasciculus, and this is likely to hamper efficient communication between perisylvian brain regions. The reported cluster on the left arcuate fasciculus was situated on the long segment, which corresponds to the classical description of the arcuate fasciculus, and connects frontal to temporal areas (Catani et al., 2005). Our finding concurs with earlier studies that reported a reduction in fractional anisotropy of the left arcuate fasciculus in dyslexia is indeed in this long segment (Gullick & Booth, 2015; Vandermosten et al., 2012). Importantly, Vandermosten and colleagues (2012) reported that this reduction is not correlated to orthographic abilities but only to speech-related abilities, such as phoneme awareness or speech-in-noise perception. In contrast to several previous studies (e.g. Feldman et al., 2010; Langer et al., 2015), we also found a significant reduction in the fractional anisotropy of the right arcuate fasciculus. Earlier studies that found this right hemisphere deficit, report that it is not, or weakly, correlated with reading and reading related abilities (Deutsch et al., 2005; Klingberg et al., 2000; Steinbrink et al., 2008). An often debated issue in the literature is the existence and nature of an hemispheric asymmetry in the temporal processing of speech. For instance, Poeppel's influential account on this topic states that both hemispheres process speech signals, but that the left hemisphere favours information in short temporal integration windows, important for phonemic

processing, whereas the right hemisphere is specialized in longer temporal integration windows, in which suprasegmental aspects of speech are processed (Poeppel, 2003). It should be noted that dyslexia is also frequently associated with deficits in these 'slower' speech processes, such as rhythm and pitch perception (Bishop-Liebler, Welch, Huss, Thomson, & Goswami, 2014; Flaugnacco et al., 2014; Goswami et al., 2002). Although speculative, deficiencies in the structural properties of the right arcuate fasciculus could be consistent with notions that individuals with dyslexia show impairments in these other aspects of speech processing (Alves, Reis, & Pinheiro, 2015; Suárez-Coalla & Cuetos, 2015).

After testing this specific hypothesis on the role of the arcuate fasciculus, we investigated the integrity of a more comprehensive model of the whole speech production system. Graph theoretical analyses on the neural connectivity in the speech network as specified by the DIVA model (Guenther et al., 2006; Tourville & Guenther, 2011) showed that, during resting-state, individuals with dyslexia have a weaker level of functional integration, as measured by the global efficiency and path length in the network. This is another indication that dyslexia might be characterized by deficiencies in the dynamics between speech perception and production. The same result was not found during speech perception or speech production. This could be due to the relatively short duration of the speech perception and production tasks. Functional connectivity was determined in a seven-minute time period for the resting state condition, while connectivity during speech perception and production was determined in data from approximately one minute. This likely affected the reliability of the connectivity estimations using this method (Birn et al., 2013).

Measuring functional activation patterns during speech perception did not reveal differences between individuals with dyslexia and typically reading controls. The literature on brain activation differences during speech perception provide mixed evidence. Perceiving single speech consonants was shown to elicit weaker activation in children with dyslexia in speech related areas (Blau et al., 2010; Žarić et al., 2014), while no differences were found in German adults during vowel perception (Steinbrink, Groth, Lachmann, & Riecker, 2012). Other studies suggest that neural differences during speech perception become clearer under adverse listening conditions (Dole et al., 2012). If dyslexia is characterized by neural under-activations during speech perception, it is probably subtle and depends on task parameters and subject characteristics. In contrast, the current study reported clear neural differences during speech production, with stronger activation during speech production tasks in both speech sensory and production areas for typical readers than individuals with dyslexia. The reported clusters included the left and right superior temporal gyrus. The superior temporal

gyrus is often associated with speech perception abilities (Chang et al., 2010; Dehaene-Lambertz, Dehaene, & Hertz-Pannier, 2002). Importantly, a number of studies suggest that activation in this core region for speech perception is inhibited during one's own speech production (Creutzfeld, Ojemann, & Lettich, 1989; Houde, Nagarajan, Sekihara, & Merzenich, 2002). An explanation for this inhibition during self-produced speech, provided by the DIVA-model, is that premotor neurons project the intended speech both to motor effectors and to speech perception cortices. As a consequence, auditory cortices could attenuate their sensitivity for this intended speech representations. In the current study, typical readers recruited these speech perception areas to a stronger extent during speech production than individuals with dyslexia. On a speculative note, individuals with dyslexia might disproportionally inhibit their speech perception areas during speech production which hinders the adequate use of auditory feedback to adjust speech production and update phonological representations. The same reasoning, although for somatosensory feedback, could be applied for the activitation differences in the left and right supramarginal and primary somatosensory gyri. These results not only show that examining speech production differences could further contribute to understanding the phonological deficit in dyslexia, but also support the hypothesis that dyslexia could be characterized by deficient speech perception-production interaction.

The results of this study suggest that dyslexia is characterized by deficient communication between areas involved in speech perception and production. Earlier literature on a phonological deficit in dyslexia focused predominantly on perceptual processes (e.g. Blau et al., 2010; Dole et al., 2012) even though speech motor components of phonological representations are also important (Guenther et al., 2006). The reported impaired structural and functional connectivity between speech areas in the current study, as well as the pattern of hypoactivations during speech production in individuals with dyslexia indicate that the communication between speech perception and production areas might be malfunctioning in dyslexia. Importantly, impairments in speech perception-production interactions almost inevitably lead to suboptimal phonological representations and this could well be expressed in deficits during speech perception or production tasks. However, to better understand the nature of the phonological deficit in dyslexia, future studies should measure the interaction between speech perception and production more directly.

The current study addresses the neurobiological basis of the phonological deficit in dyslexia from the perspective of speech perception-production interaction, and by using multiple neuroimaging techniques that test our hypothesis. At a group level, adults with dyslexia showed several impairments, both structurally and functionally, possibly associated with a reduced functioning of speech perception-production

interaction, which could relate to the origin of the phonological deficit. More specifically, speech production models suggest that using speech sensory information is crucially involved in the development and adjustments of adequate phonological representations. The results of this study provide a neurobiological basis for impaired phonological representations in dyslexia. Future studies should try to link these findings with behavioural responses on measures of speech perception-production interaction (such as response to altered auditory feedback), as well as on traditional measures of phonological and reading abilities.

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COMPETING INTERESTS

The authors declare no competing interests at the time of submission and publication.

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APPENDICES

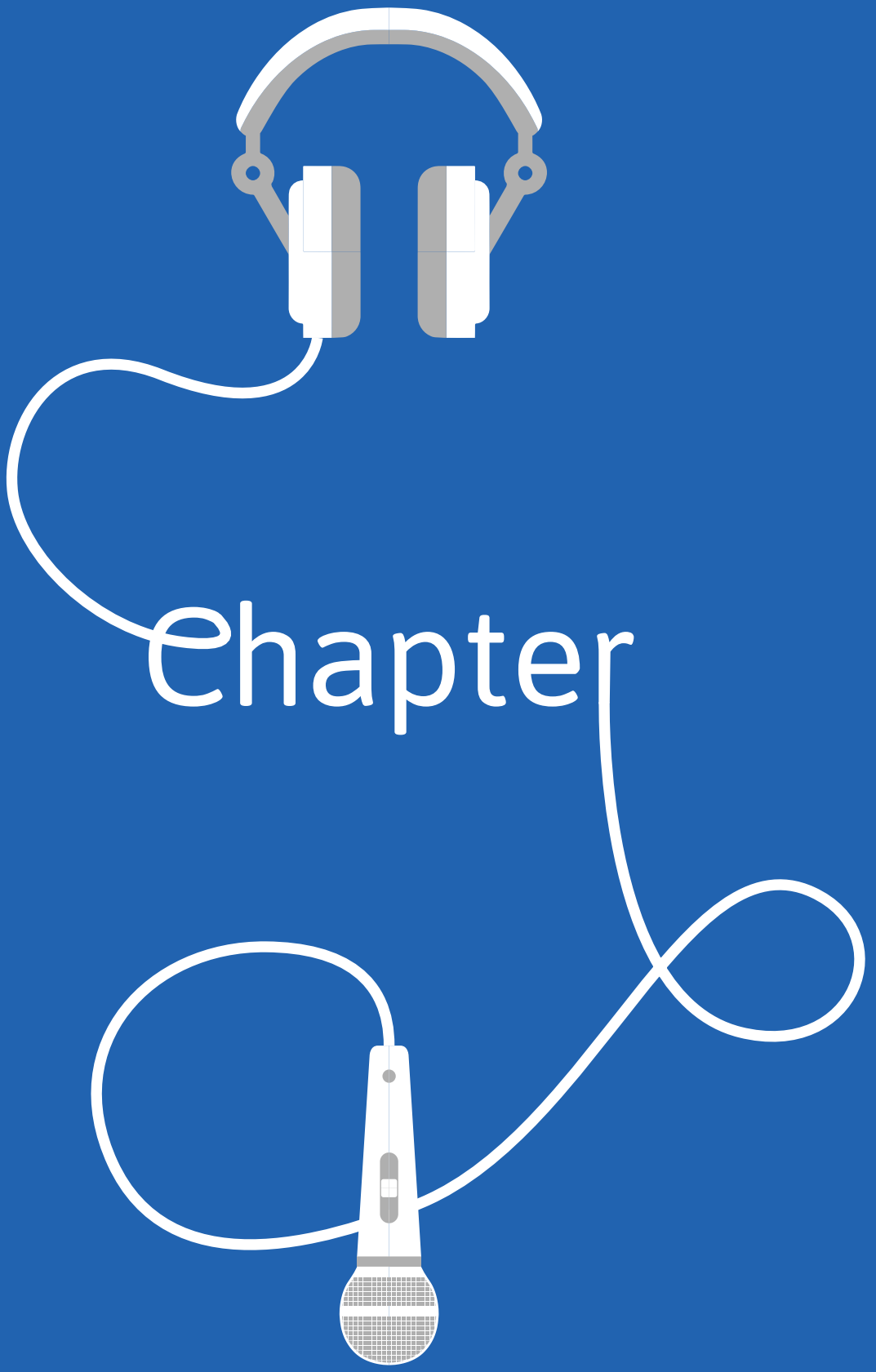
Appendix 1. The IPA transcription of the consonant-vowel stimuli used in the speech perception and production task.

be	fo	ji	lu	pu	su
by	ɣu	ju	mo	py	ty
de	ɣe	ki	my	ro	ti
do	ɦa	ko	nɛi	ry	wu
fu	ɦy	li	nu	sy	wy

Appendix 2. MNI coordinates of the ROIs of the DIVA model as used in the functional connectivity analysis;

MNI coordinates						
x	y	z	radius	modality	hemisphere	area
-36.0	-59.0	-27.0	5	motor	left	cerebellum anterior lateral
40.0	-60.0	-28.0	5	motor	right	cerebellum anterior lateral
-18.0	-59.0	-22.0	5	motor	left	cerebellum anterior paravermis
16.0	-59.0	-23.0	5	motor	right	cerebellum anterior paravermis
-10.3	-52.9	-28.5	5	motor	left	cerebellum deep cerebellar nuclei
14.4	-52.9	-29.3	5	motor	right	cerebellum deep cerebellar nuclei
-24.0	-2.0	-4.0	5	motor	left	globus pallidus
24.0	2.0	-2.0	5	motor	right	globus pallidus
-37.4	-22.5	11.8	5	sensory	left	Heschl's gyrus
39.1	-20.9	11.8	5	sensory	right	Heschl's gyrus
-56.5	14.8	4.8	5	motor	left	inferior frontal gyrus
-64.6	-33.2	13.5	5	sensory	left	posterior superior temporal gyrus
69.5	-30.7	5.2	5	sensory	right	posterior superior temporal gyrus
-59.6	-1.3	33.2	3	motor	left	primary motor: Jaw
62.1	3.9	34	5	motor	right	primary motor: Jaw
65.4	5.2	10.4	3	motor	right	primary motor: Larynx
-58.1	6.0	6.4	5	motor	left	primary motor: Larynx Extrinsic
65.4	5.2	10.4	5	motor	right	primary motor: Larynx Extrinsic
-53.0	0.0	42	5	motor	left	primary motor: Larynx Intrinsic
-56.4	0.5	42.3	5	motor	left	primary motor: Lower Lip
59.6	-3.6	40.6	3	motor	right	primary motor: lower Lip
-17.4	-26.9	73.4	5	motor	left	primary motor: respiration

23.8	-28.5	70.1	5	motor	right	primary motor: respiration
-60.2	2.1	27.5	3	motor	left	primary motor: Tongue 1
62.9	2.5	28.9	3	motor	right	primary motor: Tongue 1
-60.2	3.0	23.3	3	motor	left	primary motor: Tongue 2
66.7	2.5	4.9	5	motor	right	primary motor: Tongue 2
-60.2	4.4	19.4	3	motor	left	primary motor: Tongue 3
64.2	3.0	22.0	3	motor	right	primary motor: Tongue 3
-53.9	-3.6	47.2	3	motor	left	primary motor: Upper Lip
59.6	-7.2	42.5	3	motor	right	primary motor: Upper Lip
-59.6	-5.3	33.4	3	sensory	left	primary sensory: Jaw
62.1	-1.5	34.0	5	sensory	right	primary sensory: Jaw
-61.8	1.0	7.5	5	sensory	left	primary sensory: Larynx
-53.0	-8.0	42.0	5	sensory	left	primary sensory: Larynx
65.4	1.2	12.0	3	sensory	right	primary sensory: Larynx
53.0	-14.0	38.0	5	sensory	right	primary sensory: Larynx Intrinsic
-56.4	-5.3	42.1	5	sensory	left	primary sensory: Lower Lip
59.6	-6.9	38.2	3	sensory	right	primary sensory: Lower Lip
-58.0	-0.7	14.3	5	sensory	left	primary sensory: Palate
65.4	-0.4	21.6	5	sensory	right	primary sensory: Palate
-60.2	-2.8	27.0	3	sensory	left	primary sensory: Tongue 1
62.9	-1.5	28.9	3	sensory	right	primary sensory: Tongue 1
-60.2	-0.5	23.3	3	sensory	left	primary sensory: Tongue 2
66.7	-1.9	24.9	5	sensory	right	primary sensory: Tongue 2
-60.2	0.6	20.8	3	sensory	left	primary sensory: Tongue 3
64.2	0.1	21.7	3	sensory	right	primary sensory: Tongue 3
-53.9	-7.7	47.2	3	sensory	left	primary sensory: Upper Lip
59.6	-10.2	40.6	3	sensory	right	primary sensory: Upper Lip
-57.2	-18.4	6.9	5	sensory	left	putamen
-26	-2.0	4.0	5	motor	left	putamen
30	-14.0	4.0	5	motor	right	putamen
59.6	-15.1	6.9	5	sensory	right	putamen
-39.1	-33.2	14.3	5	sensory	left	spt
44	-30.7	15.1	5	sensory	right	spt
0.0	0.0	68.0	5	motor	left	supplementary motor area
2.0	4.0	62.0	5	motor	right	supplementary motor area
-62.1	-28.4	32.6	5	sensory	left	supramarginal gyrus
66.1	-24.4	35.2	5	sensory	right	supramarginal gyrus
-10.0	-14.0	8.0	5	motor	left	thalamus
10.0	-14.0	8.0	5	motor	right	thalamus
60.0	14.0	34.0	5	motor	right	ventral premotor cortex


Chapter

DEFICIENT RESPONSE TO ALTERED AUDITORY FEEDBACK IN DYSLEXIA

M.R. van den Bunt[1*]
M.A. Groen[1]
S. W. van der Kleij[1]
M.W. Noordenbos[2]
E. Segers[1]
K.R.Pugh[3]
L. Verhoeven[1]

[1] Behavioural Science Institute, Radboud University, Nijmegen, The Netherlands
[2] Centre for Language Studies, Radboud University, Nijmegen, The Netherlands
[3] Haskins Laboratories, Yale University, CT, New Haven, USA
* Corresponding author: Mark van den Bunt; m.vandenbunt@pwo.ru.nl; phone nr.: +31243612430; Montessorilaan 3, 6525 HR Nijmegen, The Netherlands

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ABSTRACT

Although dyslexia is characterized by a deficit in phonological representations, the nature of this deficit is debated. Neurocomputational models suggest that the quality of phonological representations depends on the integrity of sensory feedback and motor feed-forward streams. In a previous study, it was indeed shown that adults with dyslexia respond differently to online manipulations of auditory feedback, as compared to controls. In the present study, we first examined whether children with dyslexia also differ from their typically developing peers and whether individual differences within the group of the children with dyslexia are associated with the response to altered feedback. Using linear mixed-effects modeling we found that children with dyslexia, after adapting to the altered feedback, returned to their baseline to a weaker extent than typically reading peers. Furthermore, we found that children with dyslexia with poorer reading and rapid naming skills and a poorer response to intervention, adapted more strongly in response to altered feedback. In contrast, a stronger response to altered feedback was also associated with better phonological awareness skills in children with dyslexia, and this was particularly clear in those children who also had a reduced fractional anisotropy in the arcuate fasciculus. This further corroborates that speech perception-production communication in the brain is important for phonological representations and reading abilities. We propose that the current findings are consistent with the possibility of a weaker magnet of phonological representations in children with dyslexia, that might lead to a faster deviation from the category prototype under conditions of altered feedback and a weaker return to this baseline when feedback returns to normal.

INTRODUCTION

Developmental dyslexia is characterized by persistent difficulties in accurate and fluent word reading and has a neurobiological basis (Lyon, Shaywitz, & Shaywitz, 2003). One of the main deficits in dyslexia is thought to be an impairment in the quality of phonological representations. These impaired phonological representations are often hypothesized to hinder the formation of fast, stable and automatized connections between phonology and orthography (Boada & Pennington, 2006; Snowling, 1981; Sprugevica & Høien, 2003). A relatively direct way to measure phonological representations is by examining speech perception and production abilites. With respect to speech perception, individuals with dyslexia show weaker categorical perception of phonemes (Bogliotti, Serniclaes, Messaoud-Galusi, & Sprenger-Charolles, 2008) and are also reported to have hyper-sensitive within-phoneme-category perception (Serniclaes, Van Heghe, Mousty, Carré, & Sprenger-Charolles, 2004). However, some researchers did not find perception deficits in dyslexia (Law, Vandermosten, Ghesquiere, & Wouters, 2014), or argued that a phonological deficit is secondary to a general auditory deficit (Tallal, Miller, & Fitch, 1993). Others even questioned the existence of speech perception deficits and pointed to, for instance, attentional limitations in dyslexia (Ramus & Szenkovits, 2008). More recently, the phonological access hypothesis has been proposed, stating that individuals with dyslexia have adequate phonological representations but show difficulties in consciously accessing and manipulating these representations (Boets et al., 2013; Ramus & Szenkovits, 2008). In contrast to speech perception, speech production in dyslexia—a second relatively direct way to measure phonological representations—has only received scant attention. However, the studies that have been conducted suggest that individuals with dyslexia show impairments in articulatory and oral motor skills (Elbro, Borstrom, & Petersen, 1998; Malek, Amiri, Hekmati, Pirzadeh, & Gholizadeh, 2013). In summary, many studies investigated phonological deficits in dyslexia by measuring performance on metacognitive tests (e.g., phonological awareness) or by examining speech perception, with a few by probing speech production. Most of these studies point to a deficit in phonological representations, although some do not find speech perception and/or production deficits, or hypothesize that the findings are better explained by limited access to phonological representations.

In contrast, in a separate literature, work on neurocomputational models of speech motor control suggest that the quality of phonological representations hinges on the interaction between speech production (i.e. feed-forward) and speech perception (i.e. feedback) mechanisms (Tourville & Guenther, 2011). On a neural level, this interaction between speech production and sensory areas is hypothesized to be facilitated by a white matter tract that connects the involved temporoparietal (for speech perception)

and frontal (for speech production) areas: the arcuate fasciculus. The present study therefore examined the nature of the phonological deficit in children with dyslexia, by directly probing dynamic interactions between speech perception and speech production mechanisms, using both behavioural (response to altered auditory feedback) and neuroimaging (fractional anisotropy in the arcuate fasciculus) measures.

Neurocomputational models of speech perception and speech production

According to neurocomputational models of speech production, each phonological representation is associated with a feed-forward and a feedback stream (Guenther, Ghosh, & Tourville, 2006). The feed-forward stream maps the motor representations—hypothesized to be stored in the left ventral premotor cortex—of a phoneme onto the motor effectors, while feedback mechanisms (superior temporal and somatosensory/inferior parietal areas) monitor whether the output of the feed-forward trace matches the predicted auditory and somatosensory consequences (Guenther, Ghosh, & Tourville, 2006). Once a feedback monitoring mechanism detects a mismatch between the produced and intended speech production, a corrective signal is sent to the motor cortex to repair the mistake and potentially update the feed-forward representation of a certain phoneme (Houde & Nagarajan, 2011). As this feedback control is a slow and inefficient process, the feedback trace should largely disengage to optimize the computational costs of speech production once feed-forward commands are well-defined (Guenther et al., 2006). In addition, if corrective feedback signals are sent, they should be implemented slowly in the feed-forward system in order to avoid an unstable motor system (Houde & Nagarajan, 2011). These feedback mechanisms are critically involved in learning and maintaining speech abilities. For instance, deaf and hard-of-hearing children—for whom auditory feedback is not or only partially available—have significant difficulties in acquiring adequate speech production skills (Smith, 1975). Also in late adulthood, the quality of speech production is related to sensory feedback (Lane et al., 1997). With respect to the neural representations of the feedback stream, Sitek and colleagues (2013) showed that in healthy adults, the auditory cortex is sensitive to natural variations in self-produced speech and this sensory feedback is thought to be important for maintaining speech intelligibility (Niziolek & Guenther, 2013). With respect to the feedforward stream: motor, premotor and supplementary motor areas are often hypothesized to play a key role in initiating the motor production of phonemes (Guenther et al., 2006). Measuring the integrity and stability of these mechanisms in individuals with and without dyslexia may help to further understand the origin and nature of the phonological deficit.

Altered auditory feedback and dyslexia

The interaction between speech feed-forward and feedback mechanisms is often measured by an online modification of the auditory feedback someone receives while speaking (Scheerer, Jacobson, & Jones, 2016). In these experiments, participants are usually asked to repeatedly produce a syllable, while being recorded. In some instances, the auditory feedback is modified in such a way that, for example, the fundamental frequency or the frequency of the first formant is manipulated and fed back in real-time via headphones. As a result, participants hear the speech they produced, but the pitch is slightly altered or a vowel does not sound exactly as intended. The changes in speech production in response to these manipulations, reflect how speech perception is used to alter speech production. Although participants are usually not aware of the manipulation, they do typically respond by changing their speech in the opposite direction of the manipulation (Purcell & Munhall, 2006), but large individual differences exist (Lametti et al., 2012). Factors thought to influence these individual differences in the response to altered auditory feedback include: the strength of the manipulation (Niziolek & Guenther, 2013), the developmental phase of the participants (e.g., very young children do not adapt as strongly as adults; MacDonald et al., 2012; Scheerer et al., 2016), and the shape of the participants' vowel space.

Recently, an auditory feedback paradigm was used in Dutch adults with and without dyslexia to examine whether speech perception-production interactions are affected in people with dyslexia (Van den Bunt et al., 2017). In that study, participants were asked to repeatedly produce the nonword /bɛp/ while the frequency of the first formant of the /ɛ/ sound was unaltered in the baseline phase, gradually manipulated to a 25% increase during the ramp phase, held at maximum (25%) during the hold phase, and again unaltered in the after-effect phase. It should be noted that increasing the frequency of the first formant of the /ɛ/ vowel with 25% leads to a sound that is close to the English /æ/ vowel, but this sound is not phonemic in Dutch. It was found that adults with dyslexia showed a larger deviation from the baseline production in the ramp-up phase, and a weaker de-adaptation to the baseline in the after-effect phase than typically reading adults. These results were interpreted in light of the 'perceptual magnet' theory (Feldman, Griffiths, & Morgan, 2009; Kuhl, 1991), which claims that a phonetic category prototype functions as a magnet that results in relatively poorer discriminability for neighboring stimuli close to the prototype and better discriminability for stimuli that are farther away from the prototype. With respect to the response to altered auditory feedback in dyslexia, a weaker magnet could increase the response to alterations in altered auditory feedback (when the percept deviates from the phonetic category prototype) and reduce the ability to reestablish the representations when feedback is

back to normal (Van den Bunt et al., 2017). Although these findings indicate that adults with dyslexia respond differently to altered auditory feedback—which might indicate an impairment in speech feed-forward and feedback mechanisms—several issues remain: 1) To what extent is a stronger response to altered auditory feedback characteristic of children, as it was found to be of adults, with dyslexia; 2) How does the response to altered auditory feedback relate to individual differences in reading and reading-related skills.

Regarding the first issue, participants with dyslexia of the previous study were university students, who—by definition—must have found ways to compensate for their reading deficit. Therefore it is unclear whether children with dyslexia also show evidence for a weaker magnet associated with phonological representations by responding more strongly to altered auditory feedback. This is especially relevant, as feedback control is thought to be particularly important for the formation and establishment of phonological representations during childhood (Guenther et al., 2006). Regarding the second issue, an important follow-up question is how the response to altered auditory feedback relates to individual differences in reading and reading-related skills in children with dyslexia. Dyslexia is a heterogenous disorder and children with dyslexia differ in the severity and persistence of the disorder, as well as in the underlying cognitive deficits (e.g. phonological awareness, rapid naming). Reading fluency is a clear marker of the severity of dyslexia. Additionally, Lyon an colleagues (2003) argued that the lack of response to evidence-based instruction is indicative of the severity and intractability of the disorder. Administering altered auditory feedback to children from primary schools who participated in a dyslexia treatment training allows us to examine whether the persistence of dyslexia is related to the stability and/or quality of phonological representations. With respect to the associated cognitive deficits, the literature often distinguishes between phonological awareness—particularly associated with reading accuracy—and naming speed—particularly associated with reading fluency (Nelson, 2015). The precise role of these cognitive abilities is debated in a transparent orthography, in which the letter-sound couplings are highly consistent (Borgwaldt, Hellwig, & De Groot, 2005). A number of studies argue that in transparent orthographies the role of phonological awareness in reading development is relatively small (Georgiou, Parrila, & Papadopoulos, 2008; Share, 2008) and that its role further decreases over the course of development (de Jong & van der Leij, 2003). In contrast, rapid naming appears to be a stable long-term predictor of reading abilities in transparent orthographies (Furnes & Samuelsson, 2011). Relating the response to altered auditory feedback to individual differences across reading and reading-related abilities could shed more light on whether and how the interaction between speech perception and production is related to reading difficulties.

The neural basis of reading and the role of the arcuate fasciculus

Fluent reading is often related to adequate functioning of two specialized left-hemisphere networks: The dorsal, temporo-parietal, network which is classically related to phonological processing and articulation, and the ventral occipital-frontal network, which is involved in the mapping from visual representations of words onto meaning (Pugh et al., 2000). This former network is of particular importance for the interaction between speech perception and speech production. The temporo-parietal areas include the primary auditory cortex (i.e. Wernicke's area)—an area crucially involved in the perception and processing of speech input (Geschwind, 1982). Hypoactivation of these temporo-parietal areas has often been reported in people with dyslexia (Shaywitz et al., 2002). The anterior part of this network includes the (pre)motor areas and left inferior frontal gyrus and these areas are frequently reported to be involved in grapheme-to-phoneme correspondences and articulation (Long et al., 2016; Pugh et al., 2000).

These temporo-parietal and (inferior) frontal areas are interconnected by the arcuate fasciculus, which makes the arcuate fasciculus a logical choice as a tract to focus on in the context of the research questions of the current study. The arcuate fasciculus is classically thought to be involved in the sensorimotor control of speech. For instance, conduction aphasia, characterized by difficulties in speech repetition while speech perception and production as such are intact, is often related to deficiencies in the fractional anisotropy in the arcuate fasciculus (Catani & Mesulam, 2008, but see Bernal & Ardila, 2009). This is usually taken as evidence of impaired communication between the auditory cortex and speech motor areas. Importantly, an electrocorticography study showed that communication along the arcuate fasciculus is indeed bidirectional (Matsumoto et al., 2004). Communication from motor and inferior frontal areas to (auditory) sensory areas in this way concurs with the proposed neurocomputational models of speech feed-forward and feedback mechanisms (Guenther et al., 2006), in which an afferent copy of the motor commands to the articulators is sent to sensory areas to compare intended speech with the produced speech.

Additionally, the arcuate fasciculus is also one of the most frequently mentioned neural structures related to dyslexia (Andrews et al., 2010; Steinbrink et al., 2008; Vandermosten et al., 2012). Many studies have reported a reduction in fractional anisotropy, a measure of white matter organization, in the left arcuate fasciculus in people with dyslexia (Gullick & Booth, 2015; Langer et al., 2015; Vandermosten et al., 2012). However, others report a bilateral reduction of fractional anisotropy of the arcuate fasciculus (Lebel et al., 2013; Steinbrink et al., 2008), or failed to find any difference between individuals with and without dyslexia (Andrews et al., 2010; Dougherty et al., 2007; Rollins et al., 2009). The observed group differences in the arcuate fasciculus are an indication that the arcuate

fasciculus is involved in reading and/or reading-related abilities. Vandermosten and colleagues (2015) showed that individual differences in phonological skills correlated with the fractional anisotropy of several parts of the arcuate fasciclus, suggesting that this could be an underlying mechanism of how the arcuate fasciculus is related to reading skills. Relating the fractional anisotropy in the arcuate fasciculus to the response to altered auditory feedback could clarify its role in the quality of phonological representations and consequently, in reading ability.

The present study

The primary purpose of the present study was to examine how individual differences in children with dyslexia are related to the response to altered auditory feedback. However, we first examined whether children with dyslexia, when compared to typically reading peers, showed deficiencies in speech perception-production interaction. Second, we examined whether and how individual differences in the severity (i.e. reading fluency) and persistence (i.e. response to intervention) of the reading deficit and underlying cognitive deficits (phonological awareness and rapid naming) in children with dyslexia were associated with speech perception-production interaction. Third, we examined whether and how deficiencies in this perception-production interaction were associated with differences in neuroanatomy, more specifically, to differences in the arcuate fasciculus. We did so in the transparent orthography of Dutch, in which the relations between phonemes and graphemes are relatively straightforward.

The interaction between speech perception and production was measured using an altered auditory feedback paradigm, which was designed to elicit a response in all participants. For this purpose, it was decided to first measure the /ɪ/ and the /ɛ/ vowel for each participant and then set the manipulation parameters for the altered feedback individually, resulting in a complete /ɪ/ to /ɛ/ change. Based on previous research in adults (Van den Bunt et al., 2017), we hypothesized that children with dyslexia show a weaker perceptual magnet which results in a stronger response to altered auditory feedback and a weaker return to baseline when the feedback becomes unaltered again. More specifically, the following hypotheses were formulated: Firstly, a stronger adaptation was expected for the children with dyslexia, compared to typically reading controls. Secondly, within the group of children with dyslexia only, we hypothesized that stronger adaptation and weaker de-adaptation is associated with the severity and persistence of the disorder (reading ability and response to intervention, respectively) and its associated cognitive deficits (i.e. phonological awareness and rapid naming). Specifically, lower reading ability and less response to intervention, as well as poorer rapid naming and phonological awareness skills were hypothesized to be associated

with a stronger adaptation and weaker de-adaptation to altered auditory feedback. Thirdly, the hypotheses for the role of the arcuate fasciculus were harder to explicate. Conceptually, two relations in opposite directions can be envisioned between the fractional anisotropy in the arcuate fasciculus and the response to altered auditory feedback. Higher functional anisotropy of the arcuate fasciculus could facilitate the communication between speech perception and production areas and might therefore result in a stronger response to altered auditory feedback. However, adequate communication along the arcuate fasciculus could also lead to more stable feed-forward commands and hence to more reluctance to change them as auditory feedback temporarily changes, resulting in weaker response to altered auditory feedback. In light of two earlier reported findings in people with dyslexia, that (1) dyslexia is characterized by a stronger response to altered feedback (Van den Bunt et al., 2017), and (2) reports of a reduced fractional anisotropy of the arcuate fasciculus in individuals with dyslexia (Lebel & Beaulieu, 2009; Vandermosten et al., 2012), we might expect that lower fractional anisotropy is associated with a stronger response to altered feedback.

METHODS

Participants

Thirty children with dyslexia and 10 children without dyslexia were recruited to participate in an fMRI study about the neural underpinnings of response to dyslexia treatment. The data from three children with dyslexia were excluded from further analyses: in two cases the software running the key experiment (altered auditory feedback) crashed; a third participant did not comply with the task instruction to speak within the scope of the microphone. The final sample thus consisted of 27, native Dutch, children with dyslexia (M_{age} = 12.31; SD_{age} = 0.78) and 10 children with typical reading skills (M_{age} = 12.08; SD_{age} = 0.76). Neuroimaging data was available for 24 participants with dyslexia and nine children with typical reading skills. Children were recruited in several different ways: most children with dyslexia and typical readers already took part in a large longitudinal project on the evaluation of dyslexia treatment in collaboration with a dyslexia treatment provider in the Netherlands (Marant, Elst, The Netherlands) and were invited to an additional test session. Additionally, fifteen children with dyslexia were approached via the same clinical partner, but did not participate in the larger study. Finally, five typically reading children were recruited via flyers sent around to mainstream schools in the Netherlands. All parents provided active informed consent for participation of their child in the current study, as well as access to the raw reading-related scores gathered before, during and after the dyslexia treatment —in case of

children with dyslexia. The children received a small monetary gift for their participation and travel expenses were reimbursed. The study was approved by the local medical ethical committee. Participant characteristics are provided in Table 1.

Table 1. Participant characteristics

	Children without dyslexia (N=10)		Children with dyslexia (N=27)		Welch's t-test
	M ± SD	range	M ± SD	range	*t*
Age (years)	12.08 ± 0.76	11.59-13.63	12.31 ± 0.78	10.01-13.75	-.74
Reading words (correct)	93.33 ± 12.89	72-107	50.93 ± 11.30	31-75	7.51***
PA-deletion (correct)	14.33 ± 1.63	12-16	11.1 ± 2.37	6-15	4.41**
PA-spoonerisms (correct)	8.83 ± 1.94	6-11	5.52 ± 2.29	1-11	3.89**
RN-Digits (seconds)	20.33 ± 4.23	14-25	27.19 ± 5.47	20-48	-3.48**
RN-Letters (seconds)	20.66 ± 4.03	15-27	28.22 ± 5.95	21-49	-3.68**

Abbreviations: PA = Phonological Awareness; RN = Rapid Naming; ***$p < .001$, **$p < .01$

To be included in the group of dyslexic readers, participants had to have an official dyslexia diagnosis for which they underwent a standardized phonics-based treatment, available through the general healthcare system in the Netherlands at the collaborating dyslexia treatment provider. This dyslexia treatment was only available to children in whom possible comorbid disorders were not present or sufficiently under control through drug medication.

Diagnostic and intervention procedures were highly similar for all children and followed a nationally standardized protocol (Blomert, 2006). Every child that scored below the 10th percentile on reading measurements at three consecutive time-points in grade one and two was referred to a dyslexia center for an official diagnostic examination. The dyslexia diagnosis was based on a reading score of 1.5 standard deviations below average on standardized reading tests and 1.5 standard deviations below average on letter knowledge, phonological awareness or rapid naming (Blomert, 2006). If the child indeed was diagnosed with dyslexia, he/she was referred for a phonics-based dyslexia treatment. The treatment consists of 50 individual, 45-minute sessions and takes place at the school of the child. The first 12 sessions are aimed at establishing adequate letter-sound associations using primarily monosyllabic words (Tilanus, Segers, & Verhoeven, 2016). The remaining sessions are aimed at learning exception rules and speeded reading. During an intervention session children first repeated the grapheme-phoneme association or exception rule of the week before, practiced the new rule, and practiced

word reading fluency with word naming and repeated (text) reading exercises. They also received homework assignments for reading (four times 20 minutes a week) and spelling (two times ten minutes a week). Reading was assessed before, during (after 12, 36 and 48 weeks), and after treatment.

Materials

Reading ability

The ability to read words was assessed with a standardized word reading test, the *Een-Minuut-Test* [One-Minute-Test] (Brus & Voeten, 1973). This test consisted of a list of 116 printed words of increasing difficulty for which participants were asked to read as many words out loud as possible in one minute, without making any errors. The score for word reading consisted of the total number correctly read words within the time limit.

Phonological awareness

Phonological awareness (PA) was measured using two subtests of the *Dyslexie Screening Test* [Dyslexia Screening Test] (Kort et al., 2005). The first subtest was phoneme deletion in which the child was asked to repeat a word while omitting a specific sound (e.g., say *vlag* [flag] without the */v/*, answer *lag* [lay], most correct responses were nonwords). Maximum score was 16 correct items. The second subtest consisted of 11 spoonerisms (say 'Harry Potter' but switch the first sounds; e.g., 'Parry Hotter'). Having all items correct resulted in the maximum score of 11. The standardized scores of both subtests were averaged for further analyses.

Rapid automatized naming

Rapid automatized naming was measured using the letters and digit cards of the *Continue Benoemen & Woorden Lezen* [Continuous Naming and Word Reading] test (Van den Bos & Lutje Spelberg, 2014). The participant was asked to name, as fast as possible, five 10-item rows with five unique items of either letters or digits. The total time in seconds for each card was used as the score for rapid automatized naming. The standardized scores of both subtests were averaged for further analyses.

Altered Auditory Feedback

The Altered Auditory Feedback task was programmed using the Audapter software (Cai, Ghosh, Guenther, & Perkell, 2008; Tourville, Cai, & Guenther, 2013) and an external audio-card (Roland UA-25 EX, Hamamatsu, Japan). The audapter software allows to set formant adaptations for the first and second formant simultaneously. Speech productions were recorded at 48 kHz and downsampled to 16 kHz to reduce the computational load. Recording the speech signal and feeding it back occurred almost in realtime (<11 ms).

In the altered auditory feedback task, participants were first asked to say the word /bɪp/ twenty times, guided by a computer paced rhythm, once every three seconds. Then, similarly, the participant was asked to produce the word /bɛp/ twenty times. The last five /bɪp/ and /bɛp/ productions were used to determine the frequency of the first and second formants of both vowels in each participant. After this calculation, the parameters of the experiment were set individually in such a way that maximal perturbation meant a change from /bɪp/ to /bɛp/ in each participant. The baseline productions of the /ɪ/ and /ɛ/ vowel and the manipulation parameters are summarized in Table 2 and displayed in Figure 1. No significant differences in the baseline production and manipulation parameters of the experiment were found between groups.

Table 2. Overview of the baseline characteristics of the /ɪ/ and /ɛ/ production and the manipulation parameters, separately for children without and with dyslexia

	Children without dyslexia (N=10)		Children with dyslexia (N=27)		Welch's t-test
	M ± SD	range	M ± SD	range	*t*
F1 /ɪ/	285 ± 43	214-350	316 ± 59	195-461	1.75
F2 /ɪ/	2763 ± 349	2197-3251	2604 ± 197	2263-3064	1.36
F1 /ɛ/	477 ± 26	433-518	497 ± 46	426-624	1.58
F2 /ɛ/	2426 ± 255	1997-2824	2346 ± 174	2011-2811	0.91
F1 difference	192 ± 22	162-229	180 ± 53	78-275	0.96
F2 difference	337 ± 158	121-710	258 ± 73	46-413	1.52
F2-F1 difference	529 ± 163	283-878	438 ± 79	231-568	1.71

Nb. None of the t-values are significant (all p's > .05)

As illustrated in Figure 2, the altered auditory feedback experiment itself consisted of 20 baseline trials in which the feedback to the participant was not manipulated (baseline), 30 trials in which the perturbation was gradually increased to maximum (ramp-up), 50 trials in which the perturbation was held at maximum (hold), 30 trials in which the perturbation was gradually decreased (ramp-down), and finally 20 trials in which the perturbation was back to normal (after-effect). On each trial, participants were instructed to say the word /bɪp/, while the fed back signal was strongly amplified to ensure that the participants heard their voice via the headphones, rather than via air- and bone conduction. The raw and manipulated signals were saved for analyses.

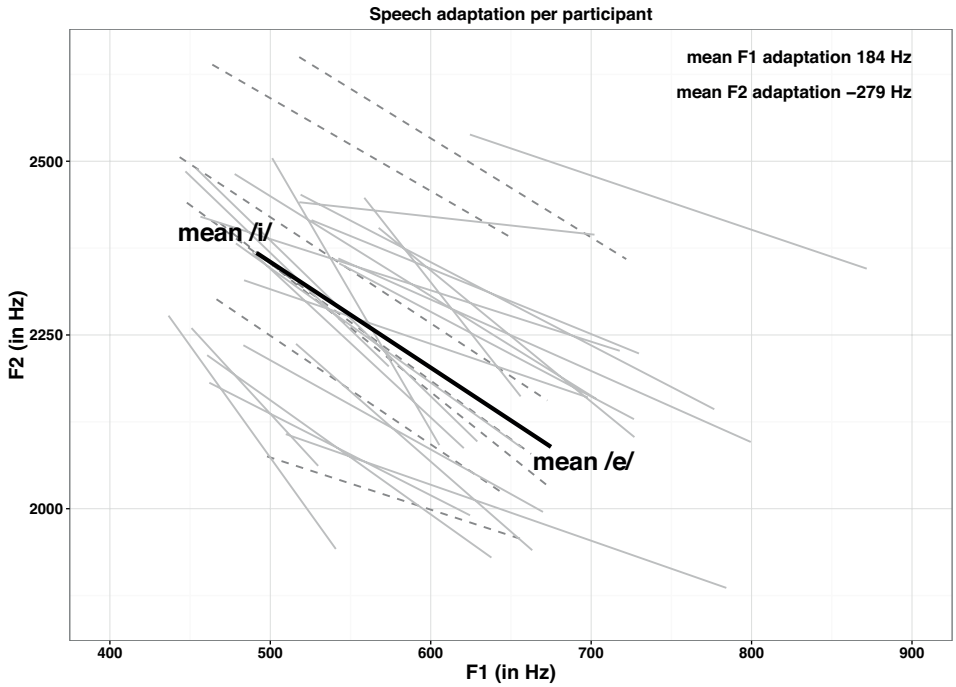

Figure 1. Speech adaptation parameters to change each participants /ɪ/ vowel into an /ɛ/ vowel under conditions of maximal perturbation. Light grey lines represent children with dyslexia; dark and dashed grey lines children with typical reading skills

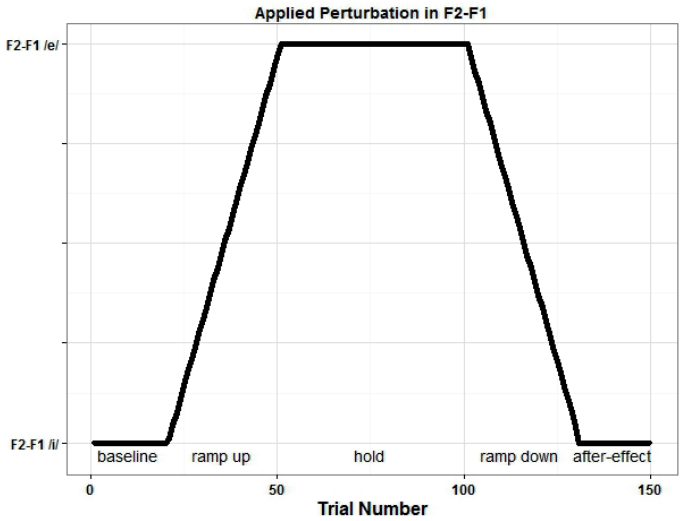

Figure 2. Overview of the different phases in the altered auditory feedback experiment.

Diffusion-weighted imaging: data acquisition and preprocessing

A diffusion-weighted imaging (DWI) scan was made using a 3T MAGNETOM Trio PRISMA^{fit} system (Siemens Healthcare, Erlangen, Germany). A 3-multiband accelerated protocol with two shells was run to obtain these images (10 unweighted images; 30 direction shell at b = 1000; 60 direction shell at b = 3000, TE = 70ms; TR = 2360ms; voxel size = 2x2x2mm^3). The resulting images were first processed using the *FSL Diffusion Toolbox* (FMRIB's Software Library; Woolrich et al., 2009). First, eddy currents were corrected using the *eddy_correct* tool. Subsequently, the brain was skull-stripped using *bet*, and a diffusion tensor model was fitted at each voxel. For six participants (1 child with typical reading abilities, five children with dyslexia), the fractional anisotropy could not be estimated due to poor tensor fitting. Each brain was then masked with the arcuate fasciculus, using the diffusion tensor imaging tractography atlas from Catani and De Schotten (2008). The mean fractional anisotropy values in the left and right arcuate fasciculus, and its subcomponents (anterior, posterior and long segment) were derived for each participant using *fslstats*.

Procedure

The child, together with the parent(s), was first invited to the dummy-scanner room in which the child could become acquainted with the MRI environment and the task in order to reduce anxiety and instruct them to lie as still as possible. After this, the child participated in the altered auditory feedback experiment while the parent signed or handed in the informed consent and filled in checklists for contraindications for participation in an MRI-study. Next, the child was placed in the real MRI-scanner for approximately 40 minutes. The MRI session started with anatomical T1 images, field map images and functional scans to map reading and speech circuits (not reported in this paper). The DWI protocol was run last. The well-being of the child was systematically monitored before entering the dummy scanner, before and throughout the scanning session in the real MRI scanner, and after the scanning session to ensure the child was happy to continue. Parents were in the control room of the MRI scanner and were able to monitor the well-being of the child as well. Reading and reading-related scores were available for all participants from the longitudinal sample or were collected after the MRI session for the other participants.

Data analyses

For each participant, the frequency of the first and second formants of the raw and adapted signals during the altered auditory feedback task were manually determined by the first author using the following procedure. The produced formants were first plotted in two ways: firstly, using linear predictive coding (LPC; Rabiner & Schafer, 1978)

in Matlab 2014a (The MathWorks Inc., Natick, MA, USA); secondly, using the default formant calculation implemented in the Audapter software (Cai, Ghosh, Guenther, & Perkell, 2008; Tourville, Cai, & Guenther, 2013). Subsequently, the author indicated the position of both formants on the y-axis if both methods overlapped. If the methods did not overlap the formant estimation of Audapter was used as default. Only if the formant estimation of Audapter was not stable, the LPC estimation was used. Because the amount of the applied F2-F1 manipulation was different for each individual, relative changes in adaptation were calculated by dividing the deviation from the mean during the baseline phase by the maximal perturbation for that participant and multiplied by 100. For instance, if someone's F2-F1 difference between the /ɪ/ and /ɛ/ vowel was 500, and if his/her baseline F2-F1 for the /ɪ/ vowel was 1500, a /bɪp/ production with F2-F1 of 1550 counted as a 10% adaptation. These relative scores were entered into linear mixed-effect models using the *lmer* function of the *lme4* package (Bates, Maechler, Bolker, & Walker, 2014) in *R* version 3.2.3 (R Development Core Team, 2015). The phase of the experiment (baseline, ramp-up, hold, ramp-down, after-effect) and the trial numbers within these phases, plus their interaction, were entered as fixed factors in the null model. A maximal random effects structure was applied as suggested by Barr, Levy, Scheepers, & Tily (2013). This means that at least random intercepts for participants, as well as by-participant slope adjustments for phase and trial were entered in the models. The best model fit was determined by performing a likelihood ratio test using the *anova* function of the *stats* package on subsequent models, starting from simply entering main effects and gradually moving to models with complex interactions. Satterthwaite approximations were used to estimate *p*-values within the model (Kuznetsova, Brockhoff, & Christensen, 2015). For all participants with dyslexia, the response to intervention was determined by calculating the growth slope during treatment using linear mixed-effects modeling: the score on word-reading was entered as dependent variable with time point during treatment as fixed factor. The random slope for each subject for this relation was used as response to intervention score.

The statistical analyses first explored the average response to the manipulation across all participants, then examined group differences between typically reading and dyslexia, and finally, individual differences within the dyslexia group only. All analyses were performed by separately entering trial number and the standardized measures of word-reading, response to intervention, phonological awareness and rapid naming into the linear mixed-effects model, after which it was examined whether 2-way or 3-way interactions with phase and trial number significantly improved the model.

Lastly, it was examined whether the fractional anisotropy of the arcuate fasciculus differed between the typically reading children and children with dyslexia with a voxel-wise statistical analysis using Tract-Based Spatial Statistics (Smith et al., 2004, 2006) and using a t-test on the mean fractional anisotropy of the total, left and right arcuate fasciculus. Next, it was examined whether the fractional anisotropy was related to individual differences in measures of word-reading, response to intervention, phonological awareness and rapid naming, and whether and how the arcuate fasciculus was related to the response to altered auditory feedback. For the reading-related measures that significantly correlated with the fractional anisotropy in the arcuate fasciculus, additional linear mixed effects models were run, in an exploratory manner, with both the behavioural measure and the arcuate fasciculus measure.

RESULTS

Response to altered auditory feedback in children with and without dyslexia

The first and second formants of 5700 /bɪp/ productions were estimated. In total, the formant calculation of 314 (5.51%) speech utterances failed due to an unstable production or estimation of one of the formants, mostly F2. On average, participants adapted their F2-F1 production with 41.41% in the direction opposite to the manipulation (range = 3.13%-91.52%). All participants changed their speech response in the direction opposite to the manipulation. The responses for both groups over the course of the experiment are depicted in Figure 3. The null model, using participants from both groups, showed that during the ramp-up phase ($\beta = 17.01$, $p < .001$), the hold phase ($\beta = 41.78$, $p < .001$), the ramp-down phase ($\beta = 32.25$, $p < .001$) and the after-effect phase ($\beta = 12.55$, $p < .001$), participants showed significant opposing responses to the applied manipulation. Moreover, a main effect for trial ($\beta = -3.60$, $p < .001$) and interaction effects of the ramp-up phase with trial number ($\beta = 13.59$, $p < .001$) and during the after-effect phase with trial number ($\beta = 3.85$, $p = .003$) were found, indicating that the adaptation response increased as a function of the trial number within these phases. Next, the factor Group (typically reading vs. dyslexia) was added to the linear mixed-effects model and compared to this null model (with a phase by trial interaction already in it). The model that was significantly better than the null model ($\chi^2(5) = 13.06$, $p = .023$), was a model with a phase by trial and a phase by group interaction. The group of children with dyslexia showed a weaker return to baseline during the ramp-down phase than the typically reading children ($\beta = 8.23$, $p = .004$).

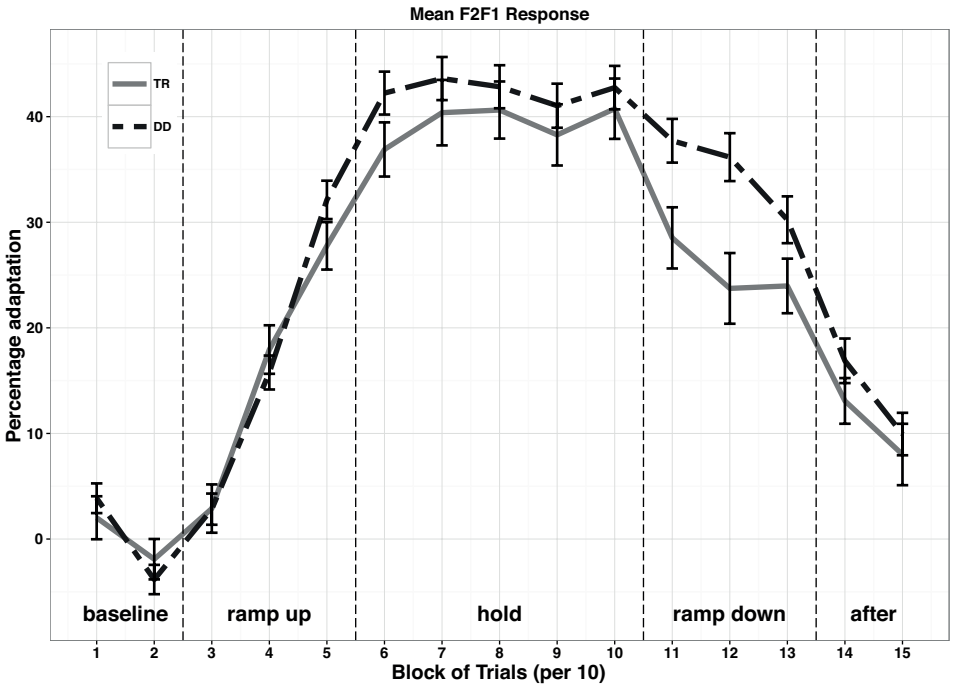

Figure 3. Mean response to the altered auditory feedback manipulation per group. TR = Typical Reading ability, solid grey line; DD = Developmental Dyslexia, dashed black line. Error bars represent one standard error of the mean.

Is the response to altered auditory feedback related to individual differences in the severity and persistence of dyslexia?

Next, it was examined within the children with dyslexia only, whether and how scores of reading and response to intervention were related to the response to altered auditory feedback. With respect to the model for reading, a model with phase by trial and phase by reading score interactions was significantly better than the null model ($\chi^2(5) = 14.10$, $p = .015$) and was not further improved by adding other interactions. In line with the group differences between typical readers and the children with dyslexia, a higher reading score among children with dyslexia was associated with weaker adaptation during the ramp-up phase ($\beta = -5.85$, $p = .042$) and stronger de-adaptation during the ramp-down phase ($\beta = -8.04$, $p < .001$). No significant differences were found for the hold and after-effect phases, however, the results were in the same direction as during the ramp-up and down phases.

Regarding the relation between altered auditory feedback and the response to intervention, convergence could not be reached using the default optimizer. The *optimx* package was used to circumvent convergence issues (Nash, Varadhan, & Grothendieck, 2013). The best model for the response to intervention included a phase by trial and a phase by response-to-intervention interaction and this was significantly better than the null model ($\chi^2(5) = 13.35$, $p = .020$). Adding interactions did not further improve the model. The only trend in the data was a weaker adaptation response during the hold phase as a function of response to intervention ($\beta = -2.47$, $p = .070$). A better response to treatment was thus associated with less adaptation as a response to altered feedback.

Is the response to altered auditory feedback related to individual differences in rapid naming and phonological awareness?

A model with phase by trial and phase by rapid naming was significantly better than the null model ($\chi^2(5) = 20.37$, $p = .001$) and adding other interactions did not further improve the model. The direction of the effects was the same as that of reading and response to intervention: A better score (i.e. faster naming) on rapid naming was associated with a weaker deviation from baseline during the ramp-up phase ($\beta = -5.01$, $p = .004$) and a stronger de-adaptation to baseline during the ramp-down phase ($\beta = -6.06$, $p < .001$). No significant differences were found during the hold phase ($\beta = -1.29$, $p = .413$) and the after-effect phase ($\beta = -2.77$, $p = .141$).

With respect to phonological awareness, a model with phase by trial and phase by phonological awareness score interactions was significantly better than the null model ($\chi^2(5) = 73.28$, $p < .001$), and adding more interactions did not further improve the model. Remarkably, a higher score on phonological awareness was associated with a marginally stronger adaptation during the ramp-up phase ($\beta = 3.60$, $p = .051$), with a stronger response during the hold phase ($\beta = 12.44$, $p < .001$) and a weaker de-adaptation in the ramp-down ($\beta = 9.51$, $p < .001$) and after-effect phase ($\beta = 8.65$, $p < .001$). The responses per phase for the individuals with dyslexia are separately plotted for individuals with scores above and below average on phonological awareness in that group in the top panel of Figure 4. Since this finding contradicts our hypothesis and also does not match the results of the relations between the response to altered auditory feedback and reading scores, we were particularly careful to assure that this finding was not driven by outliers, which was not the case.

The arcuate fasciculus and its relations with reading(-related) measures and speech perception-production interaction

Voxel-wise statistical analysis did not reveal significant clusters of decreased or increased fractional anisotropy in children with compared to children without dyslexia. Also, the mean fractional anisotropy in children with dyslexia was not significantly different from that in typically reading peers for the arcuate fasciculus as a whole ($t(18.25) = .94$, $p = .360$, $d = .440$), or for the left ($t(15.46) = .93$, $p = .369$, $d = .471$) and right arcuate fasciculus ($t(22.42) = .91$, $p = .370$, $d = .386$) separately. Correlational analyses were performed to examine the relations between the fractional anisotropy of the arcuate fasciculus and reading and reading-related measures, in children with dyslexia only. The resulting correlations are provided in Table 3. Measures of the arcuate fasciculus as a whole, or the left or right arcuate fasciculus separately only correlated significantly with scores on both measures of phonological awareness. Reading and rapid automatized naming did not correlate with the fractional anisotropy of the arcuate fasciculus.

Table 3. Correlations between the fractional anisotropy values of the arcuate fasciculus and the reading-related measures

Arcuate fasciculus	Reading	PA-deletion	PA-Spoonerism	RN-letters	RN-Digits
Whole	.07	.42*	.41*	.33	.06
Left	.09	.41*	.43*	.31	.07
Right	.05	.41*	.36*	.33	.04

Nb. *significant at p <.05; Abbreviations: PA = phonological awareness, RN = rapid automatized naming

Next, it was examined how the fractional anisotropy in the arcuate fasciculus was related to the response to the altered auditory feedback manipulation, which was only the case for the fractional anisotropy in the left arcuate fasciculus. Adding a phase by trial by left arcuate fasciculus interaction to the null model significantly improved the model ($\chi^2(10) = 23.45$, $p = .009$). The within model approximations showed that a higher fractional anisotropy was related to stronger adaptation in the ramp up and hold phase and weaker de-adaptation in the ramp down and after-effect phase. Only the weaker de-adaptation during the ramp-down phase was significant ($\beta = 5.21$, $p = .001$). Since the fractional anisotropy in the left arcuate fasciculus was significantly related to phonological awareness as well as to the response to altered auditory feedback, we examined, exploratively, whether the fractional anisotropy in the arcuate fasciulus showed interaction effects on the relation between phonological awareness and response to altered feedback.

The model with a phase by left arcuate fasciclus by phonological awareness interaction was significantly better than the model with a phase by trial and phase by phonological awareness model ($\chi^2(10) = 117.55$, $p < .001$). The approximations within the model showed that, as in the previous analysis, a higher phonological awareness was associated with a stronger deviation from baseline during the ramp-up ($\beta = 9.45$, $p < .001$) and hold phase ($\beta = 23.65$, $p < .001$) and a weaker return to baseline during the ramp-down ($\beta = 15.82$, $p < .001$) and after-effect phase ($\beta = 17.01$, $p < .001$). Having a higher fractional anisotropy in the left arcuate fasciculus was associated with a weaker deviation from baseline during the ramp ($\beta = -5.52$, $p = .006$) and hold phase ($\beta = -14.19$, $p < .001$). Also, a higher fractional anisotropy was associated with an adaptation response in the ramp-down ($\beta = -7.73$, $p < .001$) and after-effect ($\beta = -8.17$, $p < .001$) phase that was closer to baseline production. So, when controlling for the effects of phonological awareness, a higher fractional anisotropy was associated with less adaptation throughout the altered feedback experiment. Interestingly, we also found an interaction between score on phonological awareness and the fractional anisotropy of the left arcuate fasciculus for the ramp-up ($\beta = -4.55$, $p = .038$), hold ($\beta = -12.33$, $p < .001$) and after-effect phase ($\beta = -14.49$, $p < .001$). This means that the opposing pattern for the relation between phonological awareness and response to altered feedback is mainly driven by participants with a low fractional anisotropy in the left arcuate fasciculus. To illustrate the modulatory influence of the left arcuate fasciculus on the relation between phonological awareness and response to altered feedback, we plotted the correlation between phonological awareness and response to altered feedback during the hold phase for participants with a relatively weaker fractional anisotropy (below median) in the left arcuate fasciculus and with a higher fractional anisotropy in the left arcuate fasciculus in the lower panels of Figure 4.

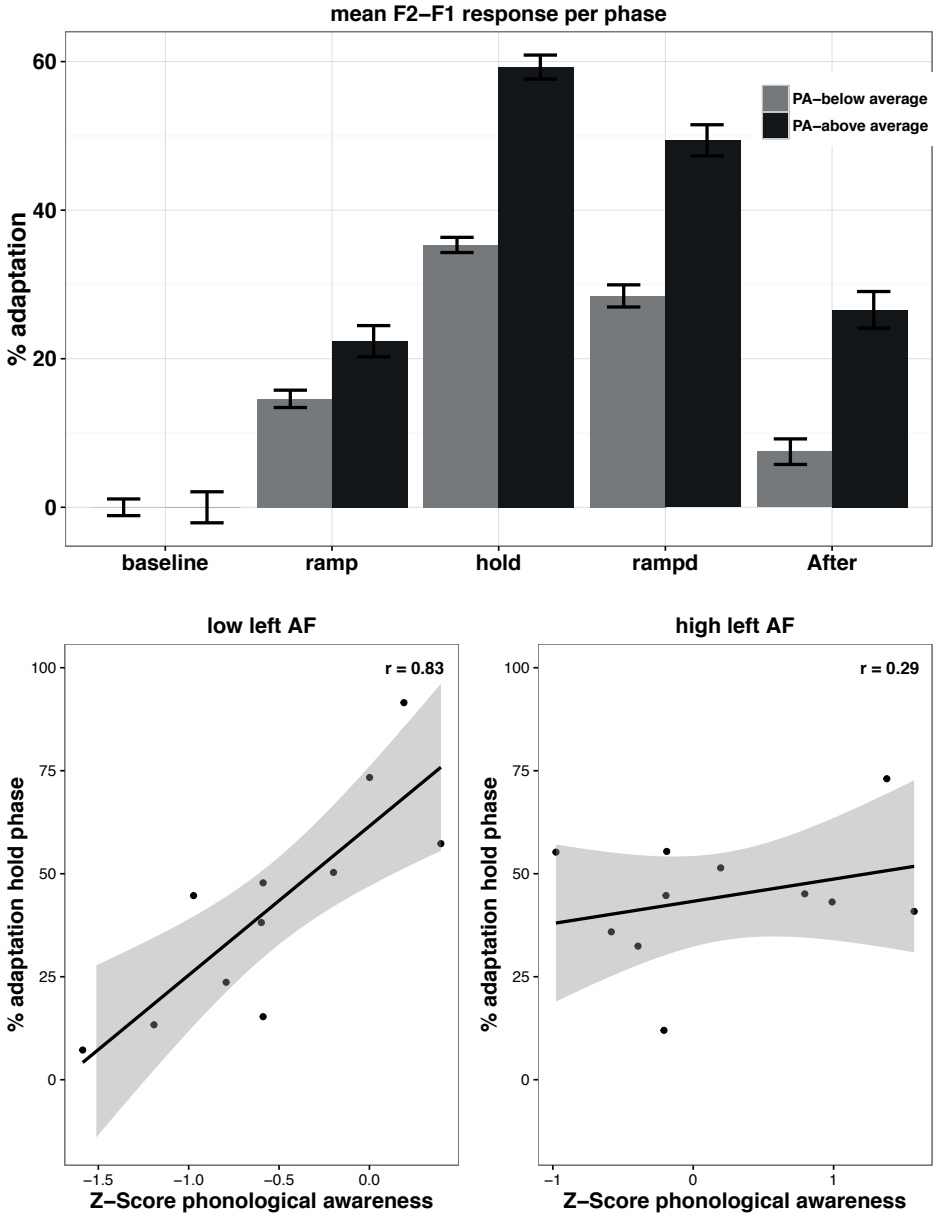

Figure 4. Mean response to the altered auditory feedback per phase for the participants with dyslexia with a phonological awareness score below and above average (top panel). Error bars represent one standard error of the mean. The bottom panels show that this finding is mainly driven by participants with a low fractional anisotropy in the left arcuate fasciculus (bottom left panel, r = .87, p = .001, 95% CI = .52-.97) and that this relation is not present for participants with a higher fractional anisotropy of the left arcuate fasciculus (r = .29, p = .381, 95% CI = -.37-.76). Shaded areas represent 95% confidence interval.

DISCUSSION

In the current study, it was first examined whether children with dyslexia responded differently to altered auditory feedback when compared to typically reading children. This group comparison showed that children with dyslexia adapted to the feedback manipulation to a similar extent, but did not de-adapt during the ramp-down phase as strongly as typically reading peers. Next, within the group of children with dyslexia only, it was examined how the response to altered auditory feedback related to individual differences in the severity of the reading difficulites and reading-related cognitive abilities. We found that more severe (word reading skills) and persistent (response to intervention) reading difficulties in children with dyslexia were associated with an impaired response to altered feedback. Similarly, lower rapid naming skills were associated with a stronger response to the alteration in auditory feedback. Contrary to our expectations, the relation between phonological awareness and the response to altered auditory feedback showed the opposite pattern. Better performance on the phonological awareness tasks was associated with a stronger response to altered feedback. Lastly, we showed that the fractional anisotropy in the left arcuate fasciculus was positively correlated with measures of phonological awareness and moderated the relation between phonological awareness and response to altered auditory feedback. The results showed that when controlled for phonological awareness skills, a lower fractional anisotropy in the left arcuate fasciculus was associated with a stronger response to altered auditory feedback. Moreover, the analyses showed that high performance on phonological awareness tasks, in combination with a weak fractional anisotropy in the left arcuate fasciculus, was associated with the strongest response to altered auditory feedback.

As noted above, children with dyslexia showed a weaker de-adaptation in the ramp down phase when compared to typically reading controls. Children with more severe and persistant reading difficulties and slower rapid naming abilities, within the group of children with dyslexia, showed a stronger adaptation during the ramp up phase and a weaker de-adaptation during the ramp down phase of the altered auditory feedback experiment. These results are in line with the observed differences in response to altered auditory feedback between adults with and without dyslexia. In that study (Van den Bunt et al., 2017), adults with dyslexia were also found to adapt more strongly in the ramp phase and to de-adapt to a weaker extent in the after-effect phase. The results of both these studies are in line with the notion that dyslexia could be characterized by a weaker magnet that causes children with dyslexia to be moved away from the category prototype more easily (under conditions of altered feedback) and be attracted back to their baseline (when feedback is unaltered again) to a smaller extent. An

important methodological difference with that study is that, in the study with adults, the amount of auditory alteration remained within a phoneme category (in Dutch), and since individuals with dyslexia are reported to exhibit better within-phoneme-category discrimination (Serniclaes et al., 2004), a stronger response to altered feedback could have been attributed to higher sensitivity to a within-phoneme-category change. The adaptation applied in the current study resulted in a complete vowel change, indicating that children with dyslexia showed this impaired response to altered feedback even when a phoneme category boundary was crossed. This renders the hypothesis that the impaired response to altered feedback results from better within-phoneme-category perception unlikely.

In short, we take the results of the current study to support the hypothesis that dyslexia is characterized by a weaker magnet. A weaker magnet might cause individuals with dyslexia to move away more easily from the prototype (non-significant in this study, but in the expected direction) and to be attracted back to the prototype when the feedback returns to normal to a smaller extent (significant in this study). In future studies, a purely perceptual measure of this magnet effect could be included to further corroborate this hypothesis. It should be noted that this weaker magnet could well have consequences for both the feedback and feedforward traces of phonological representations. A weak magnet might cause the feedback system to send error signals for relatively small deviations from the category prototype. In turn, an active feedback system could hamper the establishment of stable and reliable feedforward commands. This interpretation could relate to several earlier reported phenomena in dyslexia research. For instance, a weaker magnet could explain the increased within phoneme category perception in individuals with dyslexia, which is reported in studies on an allophonic mode of perception in dyslexia (Noordenbos, Segers, Serniclaes, Mitterer, & Verhoeven, 2012; Serniclaes et al., 2004). The earlier reported impairments in speech production in dyslexia (Catts, 1986, 1989; Foy & Mann, 2012) are also in line with the hypothesis of a weaker magnet. Specifically, Houde and Nagarajan (2011) suggested that if error signals from the feedback system are easily implemented in the forward stream, the motor control system becomes unstable. This might be the case in individuals with dyslexia.

The unexpected finding that better phonological skills within the children with dyslexia were associated with a stronger response to altered auditory feedback should be considered within the context of the Dutch orthography and national treatment procotols. Phonological awareness has been reported to be less important for reading development in transparent orthographies, such as Dutch. Some studies indeed report no relation between phonological awareness and word-reading skills (e.g. Georgiou, Parrila, & Papadopoulos, 2008), or suggest that the relation decreases during reading

development (de Jong & van der Leij, 2003). Similarly, in the current study, word reading skills, of the children with dyslexia, did not significantly correlate with phonological awareness ($r = .22$, $p = .270$). An opposite relation with the response to altered auditory feedback was, however, surprising. Although we should interpret these findings with caution, considering the small sample size, it is conceivable that relying on auditory feedback may help in developing phonological awareness skills, but may also cause a child to keep using the relatively slow phonological decoding route for word reading, rather than move towards building more efficient orthographic representations. This persistant use of the slower phonological decoding route possibly relates to the dyslexia treatment protocols that are implemented nation-wide in the Netherlands. These treatment protocols are largely based on efficacy studies in English, which is a fundamentally different language in terms of orthographic transparency (Borgwaldt et al., 2005) and, as a consequence, the treatment protocol puts a strong emphasis on mastering phonological awareness skills, before advancing to speeding up the reading process (Tilanus et al., 2016). As a result, some children receive extensive training in skills that allow them to perform better on phonological awareness measures, without a concomitant improvement in reading skills.

An important insight from the structural brain data that was included is that having a higher fractional anisotropy in the arcuate fasciculus reduced the extent to which phonological skills were associated with a stronger adaptation response. Rephrased, the increased response to altered feedback for the children with high scores on phonological awareness is particularly apparent for the children with lower fractional anisotropy in the left arcuate fasciculus. Possibly, some participants are aware of the (sub)phonemic structure of spoken language but an impaired communication between speech perception and production areas hinders the required feedback to update and stabilize feed-forward, motor, traces of phonological representations (Guenther et al., 2006). Although the arcuate fasciculus has been implicated in dyslexia and is hypothesized to relate to speech perception and/or production processes, follow-up studies should also include other white matter tracts. For instance, some studies suggest that the superior longitudinal fasciculus, rather than the arcuate fasciculus is important for the sensorimotor control of speech (Friederici & Gierhan, 2013), although it should be noted that it is not trivial to distinghuish the superior longitudinal and arcuate fasciculus in in-vivo neuroimaging. With respect to reading, whereas the arcuate fasciculus is often associated with decoding, the inferior fronto-occipital fasciculus is hypothesized to underlie whole-word recognition (Yeatman, Rauschecker & Wandell, 2013). It would be interesting to examine whether children with dyslexia with particular

difficulties in pseudoword reading (i.e. decoding) show an even stronger deviation from their baseline under conditions of altered feedback and a slower return to their baseline when the feedback is back to normal.

Although the current study provides further insight in how the interaction between speech perception and speech production is involved in reading ability in children with dyslexia, it does not provide new evidence on the etiology of dyslexia. Future studies could examine whether measures of speech perception-production interactions are prospectively predictive of early reading development. It is important to note that several authors have proposed that the phonological deficit is secondary to an underlying general auditory deficit that affects the ability to acquire adequate phonological representations (Goswami et al., 2002; Tallal, 1980). Examining the response to alterations in non-phonological forms of auditory information, such as amplitude or pitch, could further clarify the nature of the deficit in dyslexia. Moreover, it is a challenge to bring the current results in line with the recently popular view that the phonological deficit in dyslexia is an impairment in the access to, rather than the quality of, phonological representations (Ramus & Szenkovits, 2008). If anything, the results of the current study seem to suggest that phonological representations are more easily accessed and modified in dyslexia, rather than the contrary. Future studies should aim to include measures of phonological access to further disentangle these different explanations. Two methodological issues should also be addressed in future studies. First, the sample size of the individuals with dyslexia was small and subtle effects might have been missed due to a lack of power. For instance, a bigger sample size might have been able to detect whether the speed, rather than the strength, of deviating from or returning to the baseline in the altered auditory feedback task, related to individual differences in reading and reading related skills. Second, the rapid naming task relied primarily on fluency (under the assumption that accuracy was at ceiling), the phonological awareness tasks relied primarily on accuracy, and the reading task on a combination of accuracy and fluency (i.e. correctly read words per minute). In future studies it would be good to be consistent in this across tasks.

This is the first study that shows how the interaction between speech perception and production is associated with individual differences in reading and reading-related measures in children with dyslexia. It was shown that the severity and persistence of reading difficulties, and deficits in rapid naming skills, in children with dyslexia were associated with a stronger response to altered auditory feedback. We believe these findings support the notion of a weaker magnet in dyslexia, which might lead to a stronger adaptation during altered feedback and a weaker de-adaptation when feedback is back to normal. With respect to phonological awareness, we found that

better phonological skills were associated with a stronger response to altered auditory feedback, particularly for children with a low fractional anisotropy in the left arcuate fasciculus. This was attributed to the relative low importance of phonological awareness for reading in a transparent orthography, while relatively much effort is put in improving this awareness during treatment. This study further clarifies the nature of the phonological deficit and provides new opportunities for etiological research in dyslexia.

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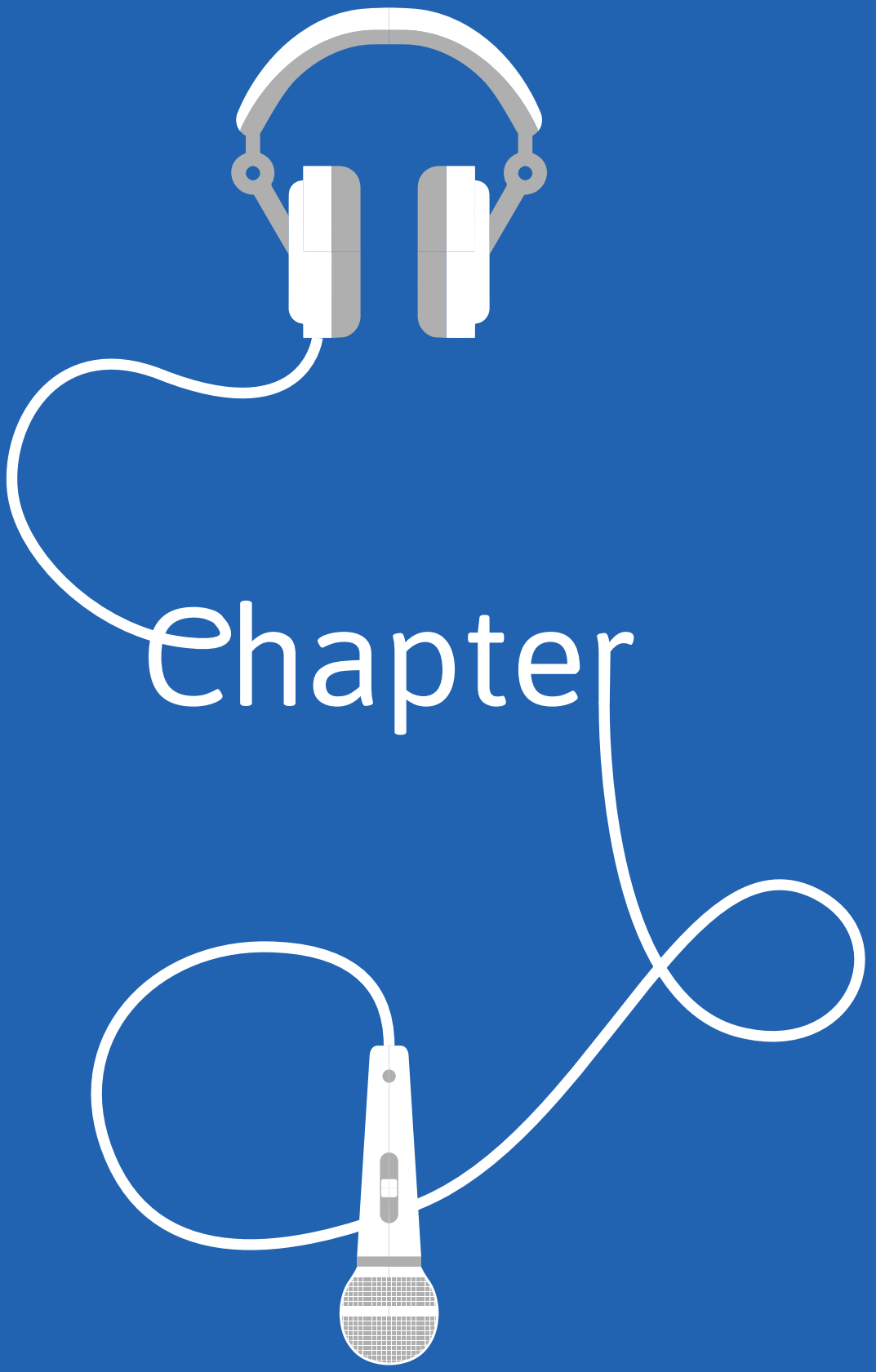
Chapter

SENSORIMOTOR CONTROL OF SPEECH AND READING DEVELOPMENT

Van den Bunt, M.R.[1]
Groen, M.A.[1]
Frost, S.J.[2]
Lau, A.[2]
Preston, J.L.[2,3]
Gracco, V.L.[2]
Pugh, K.R.[2]
Verhoeven, L.[1]

5

[1] Behavioural Science Institute, Radboud University, Nijmegen, The Netherlands
[2] Haskins Laboratories, Yale University, CT, New Haven, USA
[3] Department of Communication Sciences & Disorders, Syracuse University, Syracuse, NY USA

ABSTRACT

Instability of speech gestures such as in those with childhood apraxia of speech have been linked to literacy problems; however, studies of the role of phonological respresentations in learning to read have almost exclusively focused on speech perception. In the current study, we examined links between sensorimotor control of speech and individual differences in reading and reading related abilities in two contrastive languages, English and Dutch that vary in the regularity of their spelling-to-sound mappings. A total of 225 preliterate and literate children (77 American, 148 Dutch) between 4 to 8 years old, received an altered auditory feedback task and reading and reading-related tasks (phonological awareness and rapid naming). The altered auditory feedback design changed the perception of the first formant of the /ɛ/ vowel in the participants' production of the word /bɛp/ in four different phases: a baseline phase (no alteration), a ramp phase (gradually increasing alteration), a hold phase (maximal alteration), and an after-effect phase (no alteration again). Literate children were more responsive to alterations in auditory feedback. These children deviated from their baseline production faster than the preliterate children in the ramp phase, and had a stronger deviation from the baseline during the hold and after-effect phase. This relation was found in both languages, but reading skill was related to the response to altered auditory feedback to a stronger extent in Dutch than it was in English. Additionally, positive correlations were found between the response to altered feedback and precursors of reading in the preliterate children. We propose that these findings could be related to changes in the acoustic characteristics of the vowel space that facilitate the integration of orthography into phonetic representations. The methodological concepts and the findings of the current study open up new possibilities to examine the (reciprocal) role of phonology in orthographic learning.

INTRODUCTION

Reading skill builds on speech and language skills (Mattingly, 1972), particularly during the early stages of reading development. Unlike learning to speak, which occurs automatically, learning to read has been called an *'unnatural act'*, which requires instruction (Gough & Hillinger, 1980; Liberman, 1989) and builds on pre-existing neural networks that are developed for speech (Rueckl et al., 2015). The precise role of phonology in reading development and skilled reading (R. Frost, 1998; Leinenger, 2014) is hotly debated throughout the literature and was argued to differ across orthographies (Aro & Wimmer, 2003; Ziegler et al., 2010a, Carello, Turvey & Lukatela, 1992). For instance, theories on visual word recognition have discussed whether access to phonology is a prerequisite or an epiphenomenon during reading (Coltheart, 2003; Hardyck & Petrinovich, 1969; McCusker, Hillinger, & Bias, 1981) and whether the phonological forms that are accessed during reading resemble inner speech or are abstracted forms of phonological representations (Abramson & Goldinger, 1997; Huey, 1968). Studies on developmental dyslexia often report that impaired phonological representations underlie reading deficiencies but, again, the nature of the phonological impairment is under debate (Ramus & Szenkovits, 2008; Serniclaes, Van Heghe, Mousty, Carré, & Sprenger-Charolles, 2004). The present study probes one aspect of the speech system: integration of auditory information in the sensorimotor control of speech, in children who vary in their reading skill.

In a recent study, we examined the sensorimotor control of speech using an altered auditory feedback paradigm to probe the quality and stability of phonological representations in adults (Van den Bunt et al., 2017). We have suggested that this sensorimotor control of speech is crucially involved in the adequate development of phonological representations and directly reflects phonological processes, in contrast to measures such as phonological awareness, that also entail meta-lingusitic processing. We found that adults with dyslexia responded differently to an altered auditory feedback design that changed the first formant of the /ɛ/ vowel in the participants' production of the word /bɛp/. Adults with dyslexia adapted their speech production to a stronger extent when their productions were immediately fed back to them over the headphones with gradual alteration to the first formant of the vowel. They also returned to their baseline production to a lesser extent when the feedback was no longer altered. These findings suggest that adults with developmental dyslexia have weaker sensorimotor representations that result in both an increase in the motor response to a perceptual error and a reduction in the ability to reestablish the representation when the error is removed. In the current study, we administered this same paradigm to Dutch and American 4 to 8 year-old-children who are at different stages of reading

development (preliterate children vs. emergent readers) in order to shed new light on whether phonological representations supporting speech production and perception (and their interaction) are associated with individual or group differences in reading outcomes.

A common notion in the literature is that if the representations of the speech sounds are suboptimal, it is hard to establish stable and automatized spelling-to-sound mappings (Boada & Pennington, 2006; Elbro, Borstrom, & Petersen, 1998; Ziegler & Goswami, 2006). Indeed, the 'phonological deficit hypothesis' assumes impaired phonological representations (or access to the representations) to be at the core of difficulties in acquiring accurate and fluent reading in children with developmental dyslexia (Snowling, 1981). A considerable amount of research has investigated speech perception as a proxy for the quality of phonological representations, but results are contentious. Children with (an increased risk of) dyslexia are sometimes shown to perform worse in speech perception, or production, tasks (Brady, Shankweiler, & Mann, 1983; Hakvoort et al., 2016), and also in typically reading children, strong correlations between early speech perception and later reading abilities have been reported (Boets et al., 2011). However, perceptual deficits in individuals with developmental dyslexia have not always been found for all phonetic contrasts (Cornelissen, Hansen, Bradley, & Stein, 1996), not always under clear auditory (Ziegler, Pech-Georgel, George, & Lorenzi, 2009) or noisy conditions (Hazan, Messaoud-Galusi, & Rosen, 2012) and not for the majority of individuals with dyslexia (Manis et al., 1997). Remarkably, although phonological representations are hypothesized to have both a perceptual and an articulatory representation (Guenther, Ghosh, & Tourville, 2006), studies on the phonological deficit hypothesis in learning to read have almost exclusively focused on speech perception. The few studies that have examined speech production skills in relation to reading report that (precursors of) reading development are associated with articulation (Catts, 1997; Foy & Mann, 2001, 2012) and speaking rate (Smith, Roberts, Lambrecht-Smith, Locke, & Bennett, 2006). Evidence from clinical samples, such as childhood apraxia of speech, suggests that the stability of the speech gestures is particularly related to literacy problems as a consequence of an impaired phonological system (Lewis, Freebairn, Hansen, Iyengar, & Taylor, 2004)., Recent studies with adults have also shown that articulatory stability is associated with reading experience (Saletta, Goffman, & Brentari, 2015; Saletta, 2015). In short, not only speech perception, but also speech production appears to be related to reading ability and this is understudied in emergent readers to date. Moreover, perception and production systems are inter-dependent and feedforward/feedback processes have not been adequately examined in this population (but see Van den Bunt et al., 2017).

In the current study, the quality and stability of phonological representations are examined by probing the sensorimotor control of speech, using the same altered auditory feedback design as used in Van den Bunt et al. (2017). Studies in which altered auditory feedback is used to induce a mismatch between speech production and perception result in short-term changes in motor representations to adjust to the altered percept (Houde & Jordan, 1998; Purcell & Munhall,2006; Cai et al., 2010; Villacorta et al., 2007). More importantly, the change in production is associated with a change in perception (Lametti, Rochet-Capellan, Neufeld, Shiller, & Ostry, 2014; Shiller, Gracco, & Rvachew, 2010; Shiller, Sato, Gracco, & Baum, 2009), consistent with speech motor actions playing an important role in speech perceptual representations. As noted above, based on these perception-production studies and the notion that reading is parasitic on speech (i.e. reading uses and changes the network for speech, Mattingly, 1972; Rueckl et al., 2015), we recently explored links between the sensorimotor control of speech and reading skills in adults. Although this study showed that the response to altered auditory feedback distinguishes typical and dyslexic adult readers, an important follow-up question is how the sensorimotor control of speech is related to different stages during reading development and individual differences in typical reading development. If sensorimotor control indeed is a crucial mechanism in the development of phonological representations and if reading development hinges on the quality of phonological representations, the sensorimotor control of speech should be associated with individual differences in reading development and should also be associated with reading and readiness skills in preliterate children.

In the literature on precursors of reading development, phonological awareness, letter knowledge and rapid naming have often been used as indicators of the quality of phonological representations (Hester & Hodson, 2004; Nittrouer & Lowenstein, 2013; Snowling & Hulme, 1989; Swan & Goswami, 1997). Although these measures do reflect phonological representations to some extent, other cognitive abilities are also heavily involved in performance on these measures. For instance, phonological awareness tasks commonly involve processes that include speech perception and production, but these tasks are typically very meta-linguistic in nature and also rely strongly on attention and working memory (McBride-Chang, 1995; H. Yang, Yang, & Kang, 2014). Additionally, rapid naming of visual objects does measure the speeded access to phonological representations, but also involves sustained attention. As such, poor performance on these measures does not necessarily indicate poor phonological representations, but could also be the consequence of limited attentional or working memory capacities. Relating the sensorimotor control of speech to these classical precursors of reading will further clarify to what extent these measures reflect the quality and stability of phonological representations.

An important factor that may modulate the importance of phonological skills in learning to read is the orthographic transparency of the language in which a child is learning to read. Transparent orthographies contain mappings between spelling and pronunciation patterns that are more regular or consistent, whereas opaque orthographies have multivalent or 'inconsistent' mappings between spelling and pronunciations (Borgwaldt, Hellwig, & De Groot, 2005). Among alphabetic orthographies, English is a language with an extremely weak consistency in letter-sound mappings (Borgwaldt et al., 2005), yet the vast majority of studies on the precursors of reading has been conducted in English. This has led some to argue that the importance of phonological awareness has been overestimated and that phonological awareness is less important in transparent orthographies such as German or Dutch (Landerl & Wimmer, 2000; Share, 2008). A number of studies have directly compared the influence of phonological skills as a function of orthographic transparency, but the results are mixed. For instance, Ziegler and colleagues (2010) report that phonological awareness is a significant predictor of reading ability in all alphabetic languages, but that the relation is stronger in more opaque orthographies. Some, however, conclude that phonological awareness is only important in English (Georgiou, Parrila, & Papadopoulos, 2008; Mann & Wimmer, 2002), whereas others suggest that it is equally important across languages (Caravolas, Volín, & Hulme, 2005; Patel, Snowling, & de Jong, 2004). In a recent paper on the neurobiological foundations of literacy across different orthographies (i.e. Spanish, Hebrew, English and Chinese), it was shown that literacy acquisition was related to neural print-speech convergence independent of the orthographic transparency of the language (Rueckl et al., 2015), further corroborating the strongly intertwined relation of phonology and orthography. The same study reports that correlations between print-speech activation were slightly stronger in some limited regions in the most transparent orthography (i.e. Spanish). More cross-language contrasts of basic speech related skills, and not merely meta-phonological tasks, are required to make progress in our understanding of the cognitive basis for reading. In the current study, we examine Dutch (with a relatively transparent orthography) and English (opaque orthography).

In summary, we examine the nature and role of phonological representations for speech perception/production in early reading by examining the response to altered auditory feedback in preliterate and literate children and relating this response to individual differences in reading and reading related abilities (phonological awareness and rapid naming) across languages with contrastive orthographic depths. We target three questions. First, is being able to read associated with a different response to manipulations in auditory feedback? Because phonology and reading are strongly intertwined, we hypothesize that being able to read will be associated with a different response to alterations in auditory feedback. If learning to read indeed has an impact

on the response to altered auditory feedback, it may further elucidate what aspect of phonological representations are accessed during reading. Second, we test whether the response to altered feedback is related to individual differences in reading and reading-related abilities. On the one hand, based on previous findings of a stronger response to altered feedback in adults with developmental dyslexia (van den Bunt et al., 2017), we might expect that stronger adaptation would be associated with weaker reading and reading-related abilities. However, because learning to read also co-occurs with a change in phonological representations, a competing hypothesis might be that in typically developing children better reading and stronger reading-related abilities are associated with a stronger response to altered auditory feedback. Third, we examined whether the relations between the response to altered feedback and reading differ between languages with contrasting orthographic transparencies, English and Dutch. On the assumption that a core aspect of all orthographies entails mapping orthographic forms to the systems supporting phonology (Rueckl et al., 2015) we anticipate relations between the sensorimotor control of speech and reading in both languages; however, the strength of the relation may be stronger for transparent Dutch due to the greater covariance between spoken and written forms.

METHODS

Participants

Participants in the U.S. sample were taken from the participant pool of a three-year ongoing longitudinal study. They were recruited from the New Haven, Connecticut area over two and a half years. Children in the U.S. sample entered the study at one of two time-points: 1) between pre-school and the first half of kindergarten (N = 40; M_{age}= 5.43; SD_{age}= 0.59) or 2) between the last quarter of kindergarten and first quarter of grade 1 (N = 37; M_{age}= 7.23; SD_{age}= 0.51). These children completed the altered auditory feedback task during the second year of the larger longitudinal study (in the second half of kindergarten for the younger group: in grade 2 for the older group). All children were native English speakers with little or no exposure to a second spoken language. None of the children had any exposure to learning to read a second language. In addition to the altered auditory feedback task, all children also participated in literacy assessment sessions that measured their phonological awareness skills, rapid naming ability, letter knowledge, basic word recognition, and decoding skills. Written consent was obtained from both the primary guardians and the child participants prior to beginning the study.

Participants in the Dutch sample were recruited in two waves. During the first wave, 53 schools in the surroundings of Nijmegen were approached to participate in a study on predictors of reading development. Six schools gave permission to administer cognitive tests during school hours across three consecutive years, while children moved from kindergarten to grade 2. Response to altered auditory feedback in these children was assessed at the end of grade 2 (M_{age}= 7.98; SD_{age}= .31) in a total of 106 children. Three children were bilingual; all children were fluent speakers of Dutch. In order to examine differences in the responsivity to altered auditory feedback in children at different stages of reading development, an additional group of 43 children was recruited at one school during the second wave. Ten of them were in kindergarten (M_{age}= 5.45 years; SD_{age}= .36), sixteen at the beginning of grade 1 (M_{age}= 6.69 years; SD_{age}= .45), and sixteen at the beginning of grade 2 (M_{age}= 7.72 years; SD_{age}= .71). This school was located in a multicultural neighborhood and the majority of these children (31 out of 42) were bilingual (17 Turkish; 9 Arab; 2 Serbian; 2 Polish; 1 German; 1 Tigrinya); all these children where fluent speakers of Dutch except one, whose data was excluded from analyses. Measures of phonological awareness, letter knowledge, rapid naming and reading were obtained for all children. All parents consented with participation of their child in this study.

For this study, participants in both the U.S. and Dutch samples were classified as preliterate or literate based not on grade-level, but on the actual reading level (non-word reading score of zero is preliterate, all others are considered literate children) .

Materials

Phonological awareness

Phonological awareness was measured using the *Clinical Test Of Phonological Processing 2nd Edition* (CTOPP-2, Wagner, Torgesen, Rashotte, & Pearson, 2013) for the U.S. sample, and a translated version of the 1st Edition of the CTOPP (Wagner, Torgesen & Rashotte, 1999), by the first author, for the Dutch sample. The Elision and Blending Words subtests were administered to all participants. The Elision subtest consisted of 34 items (20 in the Dutch version), and measured the ability of an individual to repeat a word with a specific sound omitted (e.g. say /bold/ without saying /b/). The Blending Words subtest consisted of 33 items (20 in the Dutch version) and measured the ability to combine sounds to form words (e.g. What word do these sounds make: /t/ - /oy/?). Each subtest began with an age-appropriate start point to establish a basal, and ended when participants reached a ceiling indicated by the three highest consecutive incorrect responses.

Rapid Naming

Rapid naming was measured using the colors and objects cards of the *Clinical Test Of Phonological Processing 2nd Edition* (CTOPP-2, Wagner, Torgesen, Rashotte, & Pearson, 2013) for the U.S. sample and of the the *Clinical Evaluation of Language Functions 4th Edition CELF-4-NL* for the Dutch sample (Kort, Schittekatte, & Compaan, 2004). The card of colors in CTOPP-2 contained four rows by nine columns of squares filled with one of the six colors (black, green, blue, red, brown, yellow). The card of objects in CTOPP-2 contained four rows by nine columns of six objects (boat, star, pencil, chair, fish, key). Prior to beginning the rapid naming tasks, each participant had a practice session in which they named each color and object once to ensure that they are familiar with the names of the colors and objects. The Dutch cohort used a card with colors consisting of six rows with six circles filled with one of four (yellow, blue, red, green) unique colors. The card with objects consisted of six rows with six objects (circle, square, triangle or star). The total time in seconds for each card was used as the score for rapid naming.

Letter knowledge

Children in the U.S. sample completed the Woodcock Johnson Test of Achievement – 3rd Edition (WJ-III). The Letter-word identification subtest assessed participants' letter and word knowledge (Woodcock, 2001), with a total of 76 items. The subtest ranged from single letter identification (e.g. "M") to sight word identification (e.g. "therapeutic"). The task began with an age-appropriate start point to establish a basal, which was measured by the six lowest consecutive correct responses. The task ended when participants reached a ceiling indicated by six highest consecutive incorrect responses.

In the Dutch sample, letter knowledge was measured using a card with all 34 graphemes of the Dutch language on it. The child was asked to articulate all graphemes, without a time limit. The number of correctly articulated graphemes was the score on letter knowledge (both letter sounds and letter names were treated as correct). Because this measure shows ceiling effects for readers, it was only administered in the children in kindergarten or at the beginning of grade 1.

Reading

In the U.S. sample, reading ability was measured using the Tests of Word Reading efficiency (TOWRE; Torgesen, Wagner & Rashotte, 1999) non-word reading Phonetic Decoding Efficiency subtest. This test required children to read as many nonwords as possible, of increasing difficulty (in terms of length and phonological complexity), in 45 seconds.

In the Dutch sample, reading ability was measured using standardized non-word reading test *Klepel* (van den Bos, Spelberg, Scheepstra, & de Vries, 1994). This test consists of one card with 120 phonotactically legal non-words of increasing difficulty (in terms of length and phonological complexity). The children were asked to read as many items as possible within two minutes. The used reading score consisted of the total number of correctly read items in the first minute.

The total number of accurately read items was converted to a nonword per minute score for use in analyses. A log transformation was applied to the number of read nonwords per minute because the measure was skewed to the right in both languages.

Altered Auditory Feedback

The altered auditory feedback task was the same for both samples. Participants were asked to produce the non-word /bɛp/ when a specific cartoon figure appeared on the screen. The figure remained on the screen for two seconds while the participant's speech was automatically recorded at 44.1 kHz. Participants were instructed only to speak when the figure was on the screen and to say nothing other than the nonword /bɛp/ throughout the experiment. The participant's speech was amplified and accompanied by pink noise to eliminate the perception of bone-conducted sound and thus ensured that the participant perceived the altered signal instead of their own voice. The task consisted of four phases. The first phase consisted of 30 trials in which the first formant (F1) of the vowel was not altered (baseline). In the second phase, the F1 was gradually increased to the maximal perturbation of a 25 percent increase in F1 over 25 trials (ramp phase). The hold phase consisted of 25 trials in which the alteration of the feedback was kept at maximum. The after-effect phase consisted of 15 trials in which the feedback was back to normal. In the case of a missed trial (due to lack of response from the participant), the trial would be repeated immediately until a response was recorded. Thus, the set number of trials in a given phase is always reached before moving onto the next phase. A graphical depiction of the task design can be found in Figure 1. It is important to note that acoustically, the perturbation is equal across languages. On a perceptual level, however, a 25 percent increase in the frequency of the first formant of the /ɛ/ vowel results in a sound close to the English /æ/ vowel. Because this is a non-existing vowel in Dutch, perceptually, the adaptation might have been more relevant for the U.S. sample.

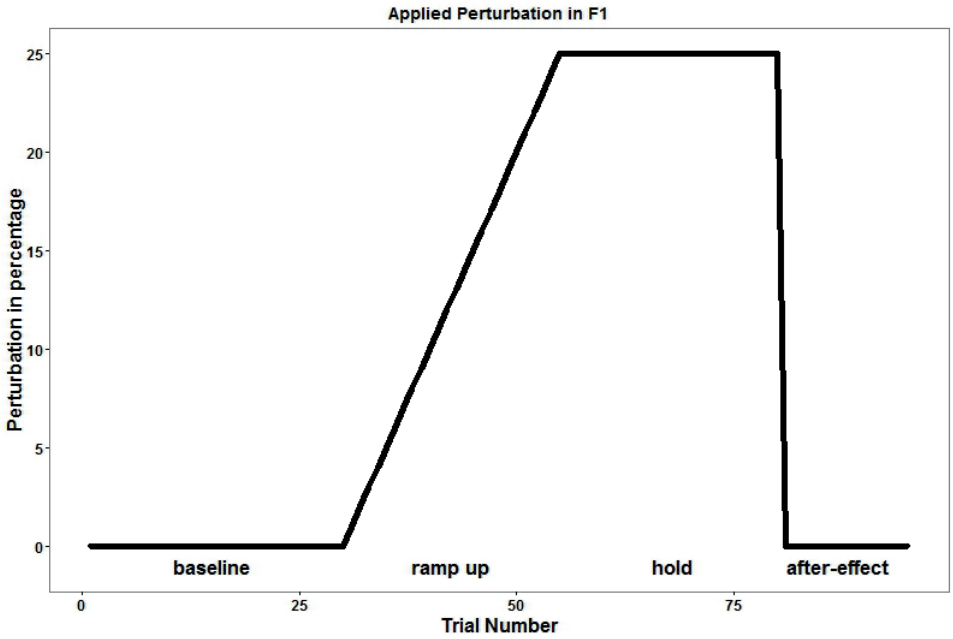

Figure 1. Design of the altered auditory feedback task.

Participants in the U.S. sample wore a lavalier micriphone around the neck. The Dutch sample spoke the nonword /bɛp/ in a Sennheiser e835 FX microphone (Sennheiser electronic GmbH & Co. KG, Wedemark, Germany) that was positioned in close proximity to the mouth. Both groups of participants wore noise-canceling headphones (HD360 Pro, Sennheiser electronic GmbH & Co., KG, Germany) to receive the (altered) feedback. Two different methodologies were used to alter the signal and present the modified feedback signal to the participants. For all U.S. participants and participants of the first wave in the Netherlands, the produced speech was amplified using a microphone amplifier (Tube UltraGain MIC100, Behringer GmbH, KG, Germany) and split in two separate streams. In one stream, the signal was unaltered. In the other stream, the formants were altered by a sound signal processor (VoiceOne; TC Helicon Vocal Technologies, Victoria, BC, Canada). Next, both streams were entered into an analogue filterbox (852, Wavetek, San Diego, CA, USA). The altered stream was low-pass filtered at 1.25 kHz, the unaltered stream was high-pass filtered at 1.25 kHz. For children of this age, the first formant of the /ɛ/ vowel is always below 1.25 kHz for children and the second formant always above 1.25 kHz. Because the alteration of the speech signal takes approximately 10 milliseconds, the unaltered stream was also delayed by 10 milliseconds using an audio delay box (DataVideo AD100; Datavideo Technologies Europe BC; Utrecht; The Netherlands).

Lastly, the two streams and the noise signal were mixed (Skytek, STM3004, Skytronic Ltd, Manchester, UK) and amplified in a headphone amplifier (HA400, Behringer GmbH, KA, Germany). For the Dutch participants recruited during the second wave (the 43 children in kindergarten, beginning of grade 1 and beginning of grade 2), the Audapter software (Cai, Ghosh, Guenther, & Perkell, 2008; Tourville, Cai, & Guenther, 2013) and an external audio-card (Roland UA-25 EX, Hamamatsu, Japan) were used to alter the speech online. To make sure the adaptation was equal across methods and samples we calculated the formants of the adapted signal and calculated whether the percentage change during the hold phase was the same for the 'Voice-One-method' in the U.S. and first wave of the Dutch sample and the 'Audapter-method' in the second wave of the Dutch sample. The adaptation during the hold phase for the Voice-One-Method was 20.59% for the U.S. sample and 19.42% for first run of the Dutch sample; the adaptation using the Audapter software was 21.10% on average. Welch's t-test did not reveal any statistically significant differences between the amount of adapation (all p's >.13).

Procedure

The experimenters explained the instructions of the altered auditory feedback task to the participants prior to turning on masking noise that eliminated the perception of bone-conducted sound. First, the participants completed a few practice trials to ensure that they understood the task and were able to speak clearly in the microphone only when the stimuli were presented. Following the practice trials, participants were reminded of the instructions one more time before beginning the experimental trials. None of the children indicated that they consciously perceived any alterations to their speech during the altered auditory feedback task. The reading and reading-related measures were collected in the same session, or within two weeks of completing the altered auditory feedback task.

Analysis

Altered auditory feedback processing

The first formants of 20,805 produced utterances and of the manipulated signal during the hold phase were estimated using a robust formant tracking algorithm that performs well in a noisy environment (such as at schools) and with speaker variability (Mustafa & Bruce, 2006). The automatic estimation of 601 (2.88%) speech utterances failed due to soft or noisy recordings. The coefficient of variation (the ratio of the standard deviation to the mean) of the /bɛp/ productions during baseline was used as a measure of speech production stability. For the response to altered auditory feedback, it was first examined whether the applied manipulation was at least greater than 10%. For six participants in the Dutch sample and four participants in the U.S. sample this did not turn out to

be the case and these participants were therefore excluded from further analyses. In addition, three participants in the U.S. sample and one participant in the Dutch sample were excluded because the first formant of a large number of trials (>25) could not be estimated.

Statistical analysis

Welch's t-tests were used to determine whether preliterate children showed more variability than literate children in the speech production during baseline. Next, it was examined whether speech production variability correlated with reading in the literate children only and with reading-related abilities, with all participants collapsed. In addition, analyses were performed across languages and for both languages separately.

Linear mixed effects models were employed on the raw F1 scores using the lmer function of the lme4 package (Bates, Maechler, Bolker, & Walker, 2014) in R (Version 3.2.3, R Core Team, 2014). The null model included a feedback phase (baseline, ramp, hold, after-effect) by trial interaction and main effects of age and language with random intercepts for subjects and random slopes for phase, and phase by trial per subject. The best model fit was obtained by likelihood ratio tests using the maximum likelihood criterion. P-values within the model were estimated using Satterthwaite approximations (Kuznetsova, Brockhoff, & Christensen, 2015). The best model fit was obtained by performing ANOVA's on sequential models, starting from the null model, gradually adding the reading-related measures and moving to complex models. The standardized F1 for each trial was calculated, for graphical purposes only, using the mean and standard deviation of the baseline phase. The random effects of the hold phase were used to get individual estimations of the response during the hold phase and this measure was used for correlational analyses.

Lastly, we examined whether reading-related measures were correlated with the response to altered auditory feedback, separately for preliterate children and literate children. Scores for phonological awareness, rapid naming and letter knowledge (for the preliterate group) were separately standardized and averaged per subtest, per language and per reading group and correlated to the slope of adaptation during the ramp phase, the total adaptation during the hold phase, and the slope during the after-effect phase, all derived from the random effects structure of the null model.

RESULTS

Motor stability and reading

The characteristics of participants included in the final sample are summarized in Table 1.

Table 1. Participant characteristics as well as descriptives for performance during the altered auditory feedback task, separately for the U.S. and Dutch preliterate and literate children.

	U.S. Sample		Dutch Sample	
	preliterate	literate	preliterate	literate
N	40	37	9	131
Age	5.43 ± .59	7.23 ± .51	5.57 ± .40	7.83 ± 53
Gender (F/M)	17/23	20/17	3/6	69/62
Reading & reading-related tasks				
Phonological awareness – Elision (U.S. Max = 34; Dutch Max =20)	11.68 ± 5.04	22.06 ± 7.11	5.22 ± 2.86	15.50 ± 3.95
Phonological awareness – Blending (U.S. max = 33; Dutch max = 20)	14.35 ± 5.61	22.53 ± 3.80	6.67 ± 3.50	17.53 ± 2.78
Letter knowledge	18.95 ± 9.60	41.08 ± 9.94	8.22 ± 5.31	N.A.
Rapid naming Objects	51.45 ± 15.02	37.87 ± 9.27	55.11 ± 7.37	38.88 ± 10.34
Rapid naming Colors	55.74 ± 22.00	40.47 ± 15.59	47.78 ± 9.07	30.88 ± 7.45
Read nonwords per minute	0	28.67 ± 17.95	0	22.07 ± 10.16
Response to altered auditory feedback-task				
Coefficient of variation during baseline	6.56 ± 2.52	5.66 ± 1.94	7.79 ± 2.35	6.25 ± 2.43
Z-score ramp phase	-.16 ± 1.16	-.19 ± 1.33	-.17 ± 1.30	-.32 ± 1.25
Z-score hold phase	-.43 ± 1.28	-.87 ± 1.63	-.32 ± 1.33	-1.07 ± 1.44
Z-score after-effect phase	-.16 ± 1.46	-.46 ± 1.49	.03 ± 1.38	-.74 ± 1.50

Nb. All scores are raw test scores unless otherwise specified.

A Welch's t-test with the variability of the first formant during the baseline phase with preliterate and literate children revealed no significant differences ($t[73.93] = 1.66$, $p = .101$). Also, no significant differences were found between preliterate and literate children for the Dutch ($t[9.21] = 1.90$, $p = .089$) and American children ($t[72.67] = 1.77$, $p = .081$), separately.

Next, we examined whether the coefficient of variation correlated with reading and reading-related abilities, separately for preliterate and literate children. The number of read non-words per minute in the literate group showed a significant correlation with the coefficient of variation ($r = -.24$, $p = .002$). All other correlations with rapid naming, letter knowledge and phonological awareness showed no significant relations with the coefficient of variation during the baseline phase (all p's > .10).

Sensorimotor control and reading

The null model for all analyses included main effects for the different phases, for trial number within each of the phases, an interaction of trial by phase, and main effects for language and age. The within model approximations revealed significant effects for age (older age was associated with lower first formant: $\beta = -35.81$; $p < .001$) and Dutch language (compared to English, $\beta = -133.18$; $p < .001$). More interestingly, significant effects were found for the ramp ($\beta = -12.46$; $p < .001$), hold ($\beta = -37.61$; $p < .001$) and after-effect phase ($\beta = -24.39$; $p < .001$), which indicate the expected opposing response to the manipulation. Additionally, significant phase by trial interaction effects were found for the ramp ($\beta = -6.64$; $p < .001$) and after effect phase ($\beta = 8.19$ $p < .001$). These interaction effects indicate that during the ramp phase, the response deviated further from baseline as a function of trial and that during the after-effect phase, the response got closer to baseline as a function of trial.

First, the ability to read (preliterate vs. literate) was added to the null model. A model with two-way interactions of phase by trial, and phase by ability to read was significantly better than the null model ($p = .003$). However, a three-way interaction of phase by trial by ability to read was significantly better than the model with the two interaction terms ($p = .006$). This model showed a significantly stronger adaptation response in the hold ($\beta = -22.51$; $p = .001$) and after-effect phase ($\beta = -24.31$; $p = .003$) for literate children. Moreover, literate children showed a significantly stronger adaptation response as a function of trial during the ramp ($\beta = -7.26$; $p = .027$) and the hold phase ($\beta = -10.89$; $p = .003$). Figure 2 depicts the response to altered feedback for the two groups with the samples of different languages collapsed because no interaction with language was found.

Next, the number of read non-words per minute, using the literate children only, was added to the null model. Adding a phase by non-words per minute interaction to the baseline model did not significantly improve the model ($p = .162$). A three-way interaction of phase by trial by non-words per minute showed a trend toward significance ($p = .017$). The within-model approximation showed that better nonword reading was associated with a faster (= trial * phase) deviation from the baseline ($\beta =$

-4.37; p = .041). Moreover, the only further significant improvement of the model is a four-way interaction with phase, trial, non-words per minute and language. The within model approximation showed that the reading-response to altered auditory feedback relation was particularly driven by the Dutch participants with a stronger (β = -20.68; p = .048) and faster (β = -16.52; p = .001) deviation from the baseline in the hold phase of the experiment. Adding other interactions did not significantly improve the model.

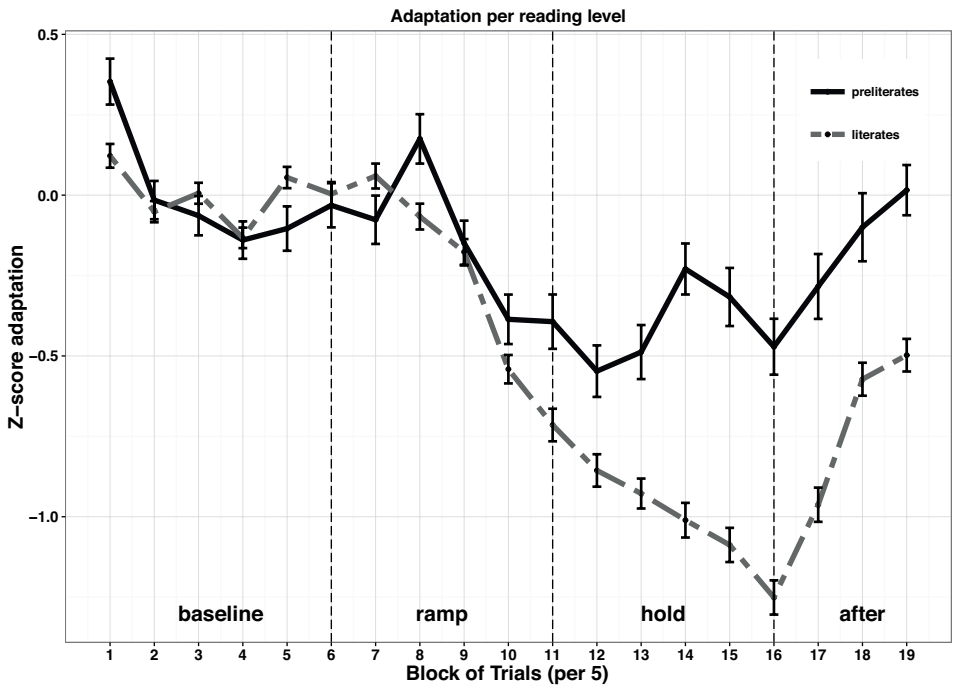

Figure 2. Adaptation response to altered auditory feedback split for preliterate and literate children; error bars represent ±1 SEM.

Lastly, correlational analyses were performed to relate the response to altered auditory feedback to reading-related measures, separately for the groups of preliterate and literate children. In the preliterate children, phonological awareness (r = -.32, p = .027), rapid naming (r = .38, p = .007) and letter knowledge (r = -.32, p = .025) all showed a significant correlation with response to altered feedback during the hold phase and no significant correlations with the slope during the ramp (phonological awareness: r = -.03; rapid naming: r = .18; letter knowledge: r = .09) or after-effect phase (phonological awareness: r = .00; rapid naming: r = -.23; letter knowledge: r = .04). For the literate

children, no significant correlations were found for phonological awareness (all r's <.05) and rapid naming (all r's <.10). Figure 3 depicts the significant correlations in the preliterate children group. Table 2 provides the correlations of the behavioral measures with the response to altered auditory feedback.

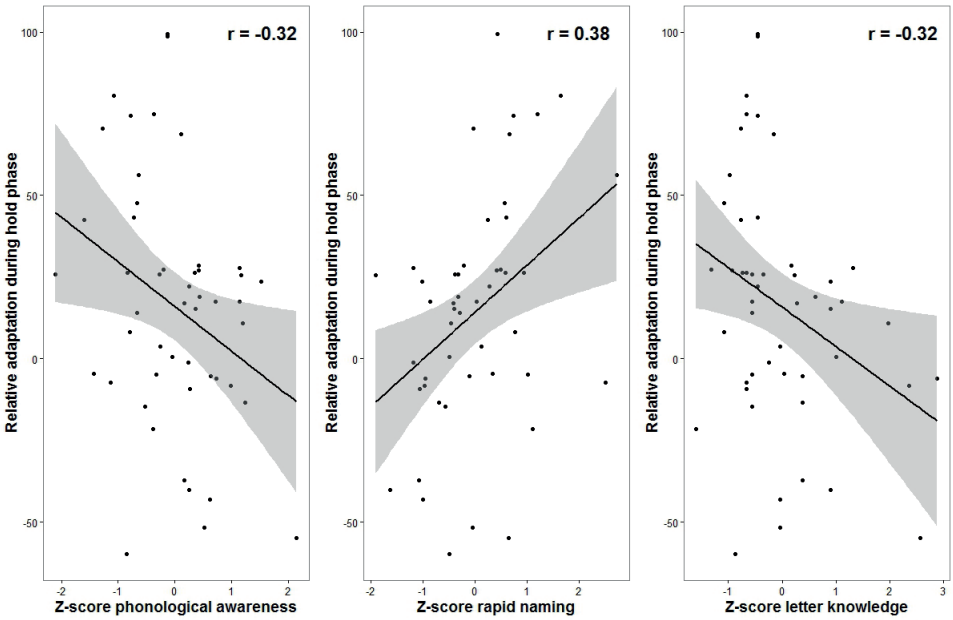

Figure 3. Significant correlations for reading related measures with response to altered feedback in the preliterate children. Scores on the y-axis represent the relative adaptation score retrieved from the random effects structure from the linear mixed effects model. The lower the score, the stronger the adaptation in response to altered feedback during the hold phase.

Table 2. Correlations of the reading related-measures with the response to altered auditory feedback. Correlations above the diagonal include the preliterate children; correlations below the diagonal include the literate children.

	1	2	3	4	5	6
1. PA	-	-.48***	.68***	-.03	-.32*	.00
2. RN	-.39***	-	-.50***	.18	.38**	-.23
3. Letter knowledge	.70***	-.65***	-	.09	-.32*	.04
4. Slope ramp phase	-.01	-.01	.07	-	.24	-.50***
5. Adaptation hold phase	-.05	.09	-.00	.46***	-	-.42**
6. Slope after-effect phase	.00	.01	.14	-.46***	-.22	-

Nb. PA = Phonological awareness; RN = Rapid naming; * p <.05, ** p <.01, *** p <.001

DISCUSSION

In the current study, we examined how the sensorimotor control of speech—hypothesized to be important for the adequate development of phonological representations—was associated with reading skill in young children. We first examined whether speech production variability was related to reading ability, which was shown within the literate children. Next, we found that the response to altered auditory feedback was related to reading stage (preliterate vs. literate) and with related to reading ability, and that it was associated with predictors of reading development in preliterate children. Moreover, and in correspondence with our hypothesis, we found that the reading-response to altered auditory feedback relation was stronger for the Dutch literate children. The literate children deviated from their baseline production faster than the preliterate children in the ramp phase, and had a stronger deviation from the baseline during the hold and after-effect phase. Within the group of literates only, better reading ability was associated with a faster deviation from baseline during the ramp phase. Lastly, the Dutch participants showed a stronger deviation from baseline during the hold phase as a function of read nonwords per minute. In the preliterate children we found that a stronger response to altered feedback was associated with better skills in phonological awareness, rapid naming and letter knowledge—important precursors to reading. These relations of the precursors of reading with response to altered feedback within the preliterate children reinforce the idea that the literacy effects (i.e. a stronger response to altered auditory feedback in the children able to read and a stronger response for the children with better reading skills) is not a general age-related effect, but is associated with learning to read itself. To sum up, after the association between the sensorimotor control of speech and reading ability that we reported in earlier work in adults, we here report that the response to altered auditory feedback is related to reading stage (preliterate vs. literate) and reading ability in children as well. Additionally, our results demonstrate positive correlations between response to altered feedback and precursors of reading in the preliterate children.

In adults, the magnitude of the response to altered feedback is influenced by the characteristics of an individual's acoustic vowel space. Specifically, Niziolek & Guenther (2013) found that perturbed feedback that results in an alteration at the border of a phonemic category yields a stronger response than altered feedback close to the center of the category (Niziolek & Guenther, 2013). A subsequent study suggests that the magnitude of the altered auditory feedback response is associated with the precision of an "efferent-evoked sensory prediction that represents a sensory goal rather than outgoing motor commands, and that is generated upstream of primary motor cortex in supplementary motor (Haggard & Whitford, 2004), or premotor (Voss, Ingram,

Haggard, & Wolpert, 2006) areas" (Niziolek, Nagarajan, & Houde, 2013, p. 16114). Hence, the response to altered auditory feedback could be indicative of one's sensory goal. With development, a number of changes occur in the acoustic characteristics of vowel productions. With increasing age, dispersion across F1-F2 space of different productions of the same vowel reduces, resulting in the area taken up by a particular vowel category becoming more compact and more separate from those of other vowel categories (Yang & Fox, 2013). If we consider the larger response to altered auditory feedback in literate compared to preliterate children in light of the developmental changes in acoustical vowel space and the findings in adults that the magnitude of the altered auditory feedback response is influenced by phonemic categories, it indicates that the exemplar areas of the vowel category in literate children is likely to have been smaller (more compact) which resulted in the alteration being closer to the category boundary. In the preliterate children, who are likely to have a vowel category that is characterized by more widely dispersed exemplars, the alteration (which was of the same size) was further away from the category boundary, presumably resulting in a smaller altered auditory feedback response.

The changes in vowel space characteristics apparently co-occur with, and possibly facilitate, the integration of orthography into previously established phonetic representations. From this perspective, learning to read is associated with changes in representations making them into highly contrastive and consciously addressable phonemic representations. Following the same line of reasoning, the positive correlation in the preliterate children between the response to altered feedback and performance on important precursors of reading is a possible indication that these hypothesized changes in vowel space characteristics precede formal literacy instruction. This interpretation matches well with neural findings in early literacy research. For instance, Pugh and colleagues (2013) report that reading readiness is positively associated with the amount of activation in inferior frontal gyrus (a region critical for speech-motor production) during the processing of spoken words and nonwords. Both the results of the current study, as well as the neural findings by Pugh and colleagues (2013), suggest an increasingly fine-grained analysis of speech as the child is shaping its (neural) speech system in order to become an efficient reader.

Interestingly, we found that reading ability was similarly related to the response to altered auditory feedback for both English and Dutch, but that reading skill was related to the response to altered auditory feedback to a stronger extent in the Dutch literate children. This finding is in line with a recent paper on the neurobiological foundations of literacy that showed that phonology and orthography are strongly intertwined in all orthographies, but that this relation is stronger in transparent orthographies (Rueckl

et al., 2015). We attribute these findings to the greater transparency of the Dutch orthography, with a stronger covariance between spoken and written forms. Due to this orthographic transparency, learning to read could have a stronger impact on phonemic representations and/or vice versa. It should be noted that the Dutch children also were older than the U.S. children and that Dutch children had relatively better decoding skills and these factors could have contributed to the stronger reading-response to altered auditory feedback relation for the Dutch children.

The methodology and the results of the current study provide many new opportunities to address the role of phonology in reading and vice versa. First, the present study shows that the relation between phonology and orthography is not only demonstrable with meta-linguistic or speech perception measures but that direct reflections of one's quality of phonological representations are also associated with reading skill. Follow-up studies could directly measure acoustic characteristics of the vowel space and relate them to reading development and the response to altered auditory feedback. Another interesting follow-up could address the issue of the direction of the association by providing literacy instruction to 4-5 years old in an experimental and longitudinal design and measure whether learning to read changes the phonological representations or whether the sensitivity to altered auditory feedback predicts learning rates in reading. On a speculative note, it is possible that, over time, reading instruction has developed to be given in a versatile period in which the vowel space is reorganizing itself and that this reorganization accommodates the acquisition of automatized and stable grapheme-phoneme representation.

The current study examined how the sensorimotor control of speech was related to reading and reading-related skills in children. The results of the study show that becoming a reader is related to a stronger response to altered auditory feedback in both a transparent and an opaque orthography. We proposed that this stronger response could be related to changes in the acoustic characteristics of the vowel space that facilitates the integration of orthographic with phonological representations. The methodological concepts and the findings of the current study open up new possibilities to examine the (reciprocal) role of phonology in reading.

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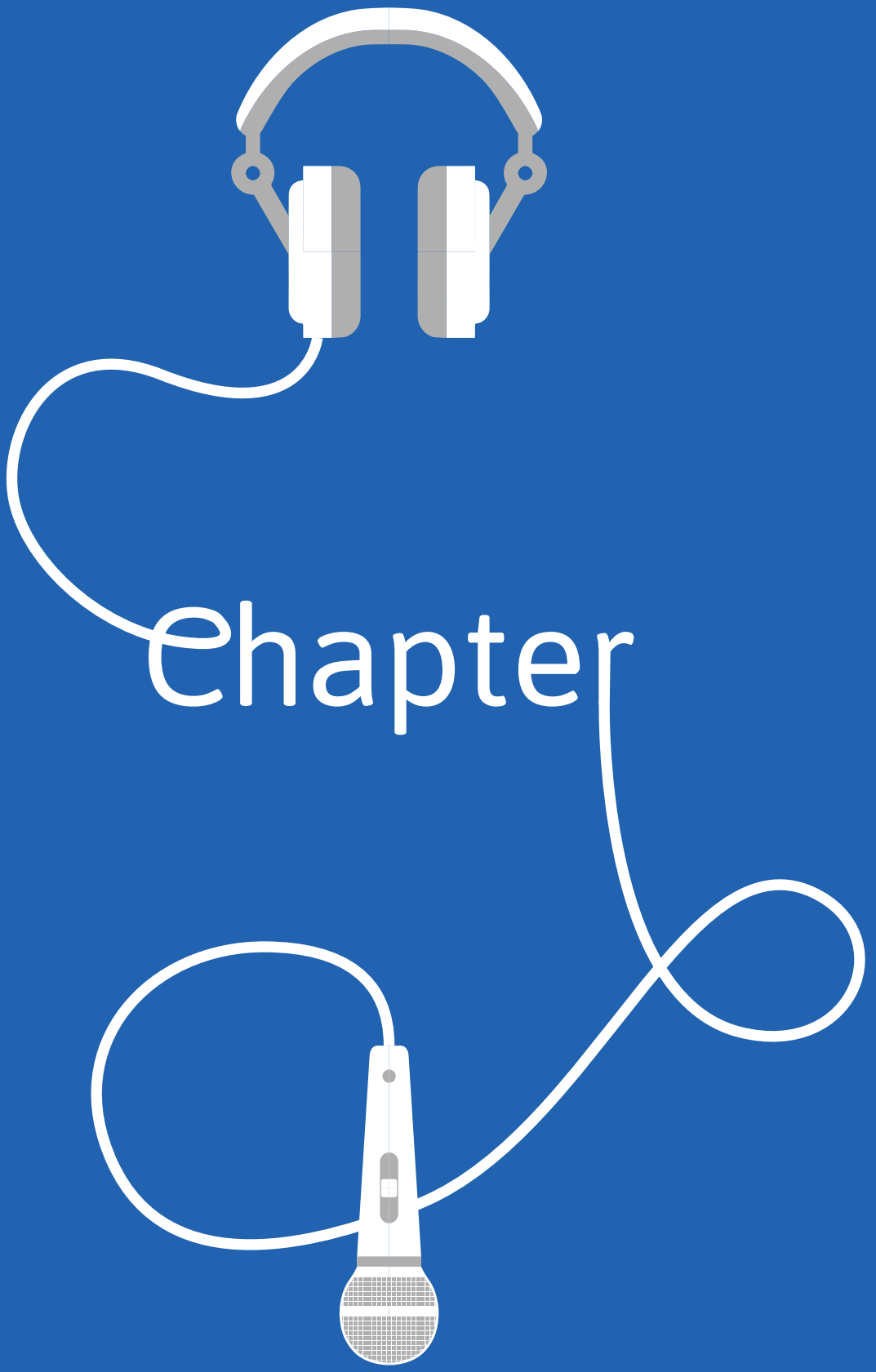
Chapter

GENERAL DISCUSSION

6

GENERAL DISCUSSION

The main conclusion that can be drawn from this dissertation is that the sensorimotor control of speech and reading skill are related. With respect to the first research question (is dyslexia characterized by an impaired sensorimotor control of speech?), I showed in Chapter 2 and Chapter 4 that on a group level, dyslexia is characterized by a stronger adaptation under conditions of altered auditory feedback, and a weaker de-adaptation when feedback is back to normal. Moreover, the differences in the methodology of the altered auditory feedback experiment (within vs. over phoneme boundary) as well as the analyses (excluding non-adapting participants vs. including all participants), as well as the age difference in the participants (adults vs. children), suggest that this finding could be a robust effect, independent of considerable differences in methodology. Additionally, Chapter 4 also showed that individual differences within children with dyslexia are related to the sensorimotor control of speech (third research question). In Chapter 3, I provided a positive answer to the second research question (is there neurobiological evidence for impaired sensorimotor control in dyslexia?). Again on a group level, dyslexia is characterized by deficient structural and functional connectivity between speech sensory and motor areas, and adults with dyslexia showed less activity in speech sensory areas when producing speech when compared to typically reading adults. Lastly, in answer to the fourth research question (how are changes in reading ability associated with sensorimotor control of speech?) I showed that being able to read is associated with a stronger response to altered auditory feedback and that precursors of reading skill were related to the strength of the response to altered feedback. These findings were interpreted in light of the dynamics of the vowel space in young children.

Although my primary research questions are answered in this thesis, many questions with respect to the implications for theory and the causal role of sensorimotor control of speech in reading development, still exist. In response to these questions, I will first describe a new perspective on the origin of dyslexia and how existing data on dyslexia fits in this new perspective. Next, I will discuss the implications of this thesis for existing theories on dyslexia and suggest follow-up studies to further support or oppose some of these theories. Lastly, I will discuss to what extent the studies in the current thesis favor a causal role of the sensorimotor control of speech in reading development and will suggest follow-up studies that would shed more light on this issue.

A sensorimotor perspective on dyslexia

The results of Chapters 2 and 4 suggest that individuals with dyslexia show impairments in the quality of phonological representations themselves, and not (only) in access to phonological representations. In these Chapters I have provided a new perspective on

the origin of dyslexia using the native language magnet theory. This theory claims that a phonetic category prototype (or center) functions as a perceptual magnet, resulting in poor discriminability of neighboring stimuli close to the prototype of the category and better discriminability for neighboring stimuli that are further away from the prototype (Feldman, Griffiths, & Morgan, 2009; Guenther & Gjaja, 1996; Kuhl, 1991). The prototype functions as if it is a magnet and 'maps' all stimuli close to the prototype onto the prototype itself. An update of the magnet theory includes a strong link between the perceptually formed representations and their associated motor commands (Kuhl et al., 2008). Articulatory movements are mapped to auditory targets and a deficient warping of the acoustic space would hinder this mapping and could affect the adequacy of both speech feedforward and feedback mechanisms. The response to altered auditory feedback has been reported to be significantly influenced by this perceptual magnet effect, with stronger adaptation responses for alterations that are further away from the category prototype (Niziolek & Guenther, 2013). The results of Chapter 2 and 4 suggest that dyslexia is characterized by a weaker sensorimotor magnet that causes individuals with dyslexia to move away from the center more easily under conditions of altered feedback (hence, stronger adaptation) and to be less attracted to the center, when the feedback is back to normal (hence, weaker de-adaptation). This weaker magnet account accommodates a multitude of earlier observed deficiencies in dyslexia such as an allophonic mode of perception (weaker magnet results in better within category discriminability close to the center) and speech production deficits (variant motor target results in more variability in speech and consequently in more speech errors). With respect to reading, a weaker magnet could also explain the difficulties in acquiring adequate grapheme-phoneme associations. If a grapheme is coupled to a phoneme with a weaker magnet, this grapheme is coupled to a more variant sensorimotor target which hampers the acquisition of efficient grapheme-phoneme associations. It is a bit harder to align the results of Chapter 5 with the perceptual magnet hypothesis of dyslexia. The results of Chapter 5 show that the strength of the response to altered feedback is positively related to (precursors) of reading. However, it should be noted that the children in Chapter 5 were, similarly to the older children and adults with dyslexia, relatively slow readers. It could be that a weak magnet accommodates the integration of graphemes and phonemes but that the phonological organization of phonemes should be further specified (hence, more reluctance to respond to altered feedback in typical readers) to allow the fast and efficient coupling of graphemes and phonemes that enables good readers to map orthography to phonology in a few hundred milliseconds.

This weaker magnet, if existing, should also be visible in 'simple' speech perception tasks, and this could be subject to follow-up investigations. For instance, one could create a vowel continuum between two vowels and ask participants with and without

dyslexia to answer which of two auditory stimuli sounded like a reference (i.e. prototype) stimulus in an AXB design. On each trial, either A or B would match the reference (the X) and the A or B is a deviant stimulus that can increase in difficulty depending on the participant's responses. Preferably, a first step would be to test everyone's vowel prototype to make sure that the reference stimulus is equivalent to the participants' own category prototype. Next, a vowel continuum between two vowels will be created, again, tailored to the participants' vowel space and after that, the test is run. Using a staircase procedure, a psychometric curve could be calculated that indicates the 'strength' of the magnet.

Implications for theories on dyslexia

In Chapter 2 and 4 I reported that (individual differences in) dyslexia is characterized by a stronger adaptation response to altered feedback and a weaker de-adaptation response when the feedback returns to normal. I take this as evidence that dyslexia is not (predominantly) associated with a deficit in phonological access (Boets et al., 2013; Ramus & Szenkovits, 2008), not with an allophonic mode of perception (Noordenbos, Segers, Serniclaes, & Verhoeven, 2013; Serniclaes et al., 2004) and not with deficiencies in statistical learning (Pavlidou, Louise Kelly, & Williams, 2010; Vicari et al., 2005).

The phonological access theory proposes that phonological deficiencies in dyslexia are attributable to the access to, rather than the quality of, phonological representations. It is claimed that individuals with dyslexia particularly show deficits in phonological processing when tasks demand conscious access, working memory and speeded responses (Ramus & Szenkovits, 2008). Impaired access to phonological representations is hypothesized to impede the fluent retrieval and manipulation of phonological representations. This theory could well explain why individuals with dyslexia perform worse on phonological awareness tasks (which requires the manipulation of representations), rapid naming (which requires speeded access) and non-word repetition (which requires verbal short-term memory). Also, responses on speech perception tasks usually require explicit and conscious responses which do require accessing phonological representations. Notably, participants of studies reported in Chapter 2, 4, and 5 were largely unconscious of the manipulation and responding did not require any speeded access and did not put any strong load on verbal short-term memory processes. Yet, individuals with dyslexia did show different responses to altered auditory feedback. If I would interpret the limited access to phonological representations very widely, without any of the constraints made by the initial proposers of the theory, I would expect that individuals with dyslexia would be less susceptible to alterations in auditory feedback, while the opposite was found. The alternative explanation of impaired phonological

access should encourage research with dyslexia to measure phonology directly if the hypotheses relate to phonological deficits, rather than measuring meta-knowledge of phonology, which is the case in tasks like phonological awareness.

The theory of an allophonic mode of speech perception in dyslexia suggests that the phonological deficit in dyslexia arises from better discrimination of allophonic differences of a phonemic category, whereas typical readers show more evidence for categorical perception (Serniclaes et al., 2004). This sensitivity for within-category variation could cause weak or absent phoneme-level representations, which are important to develop stable and automatized grapheme-phoneme correspondences. The perturbation described in Chapter 2 remained within a phonemic category, while in Chapter 4, the perturbation crossed a phoneme category. The stronger response to altered feedback in the ramp phase in Chapter 2 is the only result that possibly fits with the theory of an allophonic mode of perception. An allophonic mode of perception could have lead the individuals with dyslexia to be (unconsciously) more sensitive to the manipulation and therefore respond to a stronger extent. However, the weaker de-adaptation in Chapter 2 in the after-effect phase, as well as the results of Chapter 4 are not in line with this theory. Instead, the strongest differences between individuals with dyslexia and typical reading abilities were found when the feedback is returning to normal and individuals with dyslexia do return to their baseline to a lesser extent under these conditions. If dyslexia indeed is characterized by an allophonic mode of perception, both a stronger adaptation and a stronger de-adaptation would be expected.

The theory of statistical learning deficits in dyslexia proposes that dyslexia is characterized by impaired statistical learning. Statistical learning is hypothesized to be involved in learning to read by extracting statistical patterns from speech and print and to use this knowledge to learn, for instance, grapheme-phoneme correspondences (Pacton, Fayol, & Perruchet, 2005). The altered auditory feedback paradigm could also be viewed as a task that examines one's ability to extract and detect acoustic (ir)regularities and subsequently adjust the speech production signal to compensate for the detected irregularities. As such, impaired statistical learning would suggest a weaker/slower adaptation under conditions of altered auditory feedback and weaker/slower de-adaptation when the feedback returns to normal. Again, the complete pattern of results is not suggesting that this is the case. Participants with dyslexia appear to deviate from their baseline to a stronger extent under conditions of altered auditory feedback. Only the weaker de-adaptation when the feedback returns to normal would fit this theory.

The studies described in this thesis appear to support the phonological deficit hypothesis, which states that dyslexia is characterized by impaired phonological representations, however, this theory is not specific to the underlying cause that might lead to deficient representations. The studies described in the current dissertation are not directly addressing the role of general auditory processing, cerebellar or magnocellular deficits (see follow-up suggestions). Still, I think it is justified to put forward the idea that dyslexia is characterized by a weaker sensorimotor magnet, which integrates the results of the studies in this thesis.

Theories of dyslexia: Directions for future research

The studies described in this dissertation are the first that relate the sensorimotor control of speech to reading skill and many questions need to be addressed in follow-up research.

First, the sensorimotor control examined in the studies described in this dissertation manipulated a phoneme only, which leaves the specificity of the reported deficits in dyslexia open to debate. For instance, it is still possible that dyslexia is characterized by a general auditory processing (Goswami, 2015) deficit that also impacts speech processing. Therefore, a follow-up study could include measures of the sensorimotor control of phonology and general auditory stimuli. More specifically, adding a pitch altered auditory feedback paradigm could elucidate whether the phonological deficit is specific to phonological representations (i.e. impaired response to formant adaptation study, not to pitch adaptation) or non-specific to phonological representations (i.e. equivalent response to formant and pitch adaptation). Pitch adaptation studies commonly use similar paradigms and evoke similar responses (adapting the pitch in opposing direction of the manipulation) as formant adaptation studies (Burnett, Freedland, Larson, & Hain, 1998; Scheerer, Jacobson, & Jones, 2016) and would hence be an ideal comparison to further clarify the nature of the phonological deficit in dyslexia. Additionally, the magnocellular (Galaburda & Livingstone, 1993; Stein, 2001) and cerebellar (Nicolson, Fawcett, & Dean, 2001) theories on dyslexia suggest that the underlying mechanism responsible for the deficits in dyslexia is not even specific to the auditory modality, but is more a general sensory problem. Adding measures of the sensorimotor control of vision could further support or dismiss these general sensory theories.

The second follow-up suggestion relates to measuring neural signals when individuals with and without dyslexia participate in an altered auditory feedback task. In Chapter 3, I related structural and functional neuroimaging measures of brain areas hypothesized to be involved in the sensorimotor control of speech and found differences between

the group of individuals with dyslexia compared to the typical readers. However, administering altered auditory feedback in the scanner could examine the brain systems in action that are involved in this sensorimotor control. The outcomes of such a neuroimaging study could be used to understand what exactly is impaired in dyslexia in the sensorimotor control of speech. For instance, if the only difference between individuals with a without dyslexia would be a very local hypo- or hyperactivation of brain areas thought to be involved in speech perception or production, this could indicate that the primary problem in dyslexia is perceptual or productional in nature. Alternatively, if the main difference is found in cerebellar areas (also implicated in the neurobiology of sensorimotor control (Guenther et al., 2006), this could be seen as further support for the cerebellar theory on dyslexia.

Sensorimotor control of speech and reading: cause or consequence

The studies described in this dissertation do, in my opinion, convincingly show that the sensorimotor control of speech is associated with reading and reading development. Theoretically, altered auditory feedback paradigms tap both into the mechanisms that are responsible for developing adequate phonological representations, as well as measuring the quality and stability of the phonological representations themselves. Since the vowels that I adapted in all these studies are existing, already established vowels in all participants (although, to a lesser extent in children), I tend to believe that the reported associations between reading and response to altered auditory feedback reflect differences in the quality and/or stability of phonological representations, rather than differences in the underlying mechanisms responsible for the development of these representations. This idea is particularly based on the following line of reasoning: Adequate development of phonological representation is thought to hinge on the interaction between speech feedback and feedforward mechanisms (Hickok et al., 2011; Houde & Nagarajan, 2011). Specifically, the feedforward mechanism maps the phonological representation onto motor effectors while the feedback mechanism monitors whether the produced (sensory) output matches the intended sensory goals. In case the feedback mechanism systematically detects the same error, the feedforward trace will be updated to increase speech accuracy and reduce computational costs (Guenther et al., 2006; Hickok et al., 2011). Furthermore, feedback control is hypothesized to be particularly important in the early phases of speech development to facilitate the development of adequate representations, and this control should diminish with age to reduce these computational costs of speech production and increase fluency (Guenther et al., 2006). Therefore, if the altered auditory feedback paradigm would predominantly measure this process of adjusting existing phonological representations, which is hypothesized to be particularly important in young children, it is reasonable to expect

the strongest response to altered feedback in the youngest participants of the studies described in this thesis. However, in my studies, the only group that hardly responded to manipulations in altered auditory feedback was the youngest group of participants (i.e. the preliterate children).

In short, I conclude that the response to altered feedback possibly did not predominantly reflect the underlying mechanism responsible for the quality of phonological representations, but instead, reflected the quality of phonological representations themselves. This conclusion does not imply that sensorimotor control of speech is not responsible for the underlying mechanism that results in deficient phonological representations in dyslexia, it only indicates that I probably did not measure it. Hence, it remains an open question whether the quality of phonological representations is causally related to reading skill (Boada & Pennington, 2006; Elbro, 1998). It is beyond the scope of current discussion to discuss the literature on this topic extensively, but that literature is inconclusive. More specifically, training phonological skills has been reported to foster reading development (Brady, Fowler, Stone, & Winbury, 1994; Bus & van IJzendoorn, 1999), but learning to read, in turn, could also impact phonological development. For instance, illiterate adults score lower on a test where initial phonemes should be detected (Morais, Bertelson, Cary, & Alegria, 1986), have less precise phonemic categories (Serniclaes, Ventura, Morais, & Kolinsky, 2005) and are worse in manipulating speech sounds (Read, Yun-Fei, Hong-Yin, & Bao-Qing, 1986). In addition, exposure to orthography, which is usually less in individuals with dyslexia, also has an impact on speech production (Saletta, Goffman, & Brentari, 2015; Saletta, 2015). With respect to the studies described in this dissertation, strong claims on the causality of the phonology-orthography relation are hard to make. A few remarks can be made however. First, the correlations in Chapter 5 in the preliterate children between important precursors of reading and the response to altered auditory feedback suggest that sensorimotor control of speech is important prior to formal literacy instruction. Second, the opposing directions of the response to altered feedback with reading relation in Chapter 5 (i.e. stronger response associated with being able to read) and Chapter 2 and 4 (i.e. stronger response associated with reading impairments), suggest an age by reading by sensorimotor control interaction that, although speculative, could imply different roles of the sensorimotor control of speech throughout development. For instance, it is possible that differences in the organization of the vowel space could accommodate the binding of graphemes with phonemes (hence, stronger response to altered feedback) while the same vowel space organization hampers the stability and fluency of these phoneme-grapheme correspondences (hence, stronger response in dyslexia).

Cause or consequence: follow-up studies

This issue of causality could be addressed by a multitude of different follow-up studies, of which I would like to suggest a few. An obvious suggestion is following children, before, while and 'after' they develop reading skills in a longitudinal design in which at each timepoint, the sensorimotor control of speech is also measured. This design would allow for measuring whether the sensorimotor control of speech is predicting reading development, how learning to read changes phonological representations and how the role of sensorimotor control of speech might be different for beginning and advanced readers. An important note that should be taken into account is the phonological organization of the very young, prereading, children. The phonological organization of speech is often shown to become increasingly segmentalized with age (Ferguson & Farwell, 1975; Fowler, 1991) and it could be that single phoneme representations do not exist in these young children. As a consequence, a formant-adaptation study that alters the vowel only in a typical consonant-vowel-consonant utterance, could change only part of an existing representation in young children, while changing a complete representation in older children. Administering an experimental design that, for instance, affects the fundamental frequency of speech could bypass this issue while still measuring speech related sensorimotor control.

The suggested longitudinal design has many advantages, but is very time-consuming and there could be a need for ongoing methodological changes depending on the data. An alternative approach to estimate the causal role of the sensorimotor control of speech in reading development is by including children at risk for dyslexia. These children, as a group, will perform more poorly on reading measures later in life, while not having been exposed to formal literacy instruction yet. If children at-risk for dyslexia show a different response to altered feedback when compared to children not at-risk for dyslexia, this would suggest a causal role of sensorimotor control in reading development.

Lastly, a follow-up study could also examine the learning of non-native sounds using the sensorimotor control of speech framework. As mentioned before, because the studies in this dissertation alter existing sounds it is difficult to untangle whether the response to altered auditory feedback reflects the underlying mechanism involved in sound learning or whether it reflects the quality of phonological representations themselves. Although it would be optimal to examine these underlying mechanisms in developing children as well, a first step could be to investigate whether adults with and without dyslexia differ in their learning rates of non-native sounds. I would like to make clear that this is different from examining whether individuals with and without dyslexia differ in non-native speech sound discrimination, which has been suggested (Serniclaes, Van

Heghe, Mousty, Carré, & Sprenger-Charolles, 2004) and contradicted (Soroli, Szenkovits, & Ramus, 2010). My suggestion is to examine whether the development of new speech sound categories and their integration in the vowel space is related with reading skill.

To summarize, based on models of speech production, I would expect the sensorimotor control of speech to be causally related to reading and reading development, however, the empirical data described in this dissertation and throughout the literature are still inconclusive. Follow-up studies are needed to clarify the causal role of sensorimotor control of speech in reading development.

FINAL CONCLUSION

In this dissertation, I provided evidence that the sensorimotor control of speech is associated with reading skill. Adults with dyslexia showed a stronger adaptation response to altered auditory feedback and a weaker de-adaptation response when the feedback was back to normal. Individual differences in children with dyslexia showed a similar pattern with a stronger adaptation and weaker de-adaption that was associated with poorer word reading skills. Neuroimaging measures further supported that dyslexia could be related to a deficient speech perception-production interaction. The results of the presented thesis showed that dyslexia is characterized by impaired phonological representations, and not (only) impaired access to phonological representations. I proposed that these results fit best with the notion that dyslexia is characterized by a weak perceptual magnet which makes it easier to be moved away from baseline, and harder to return. The altered auditory feedback paradigm allows to directly compare contradicting theories of dyslexia and provides an interesting and thought provoking new window for future dyslexia research. Also, the finding that learning to read is also associated with a stronger response to altered auditory feedback suggests that the role of sensorimotor control of speech changes during development.

Considering the importance of literacy skills for an individual's academic and economic prospects as well as the societal costs associated with low literacy skills, there is probably no need to convince anyone of the relevance of research into the origins of differences in reading skill. It should be noted that, considering the many questions that this thesis is generating, the results of this thesis are not directly applicable to improve reading skills of clinical or general populations. However, I am convinced that the main interpretation (weaker magnet in dyslexia) and/or the used methodologies (measuring sensorimotor control) will help the field move forward which could ultimately result in advancements in dyslexia prevention, assessment and intervention.

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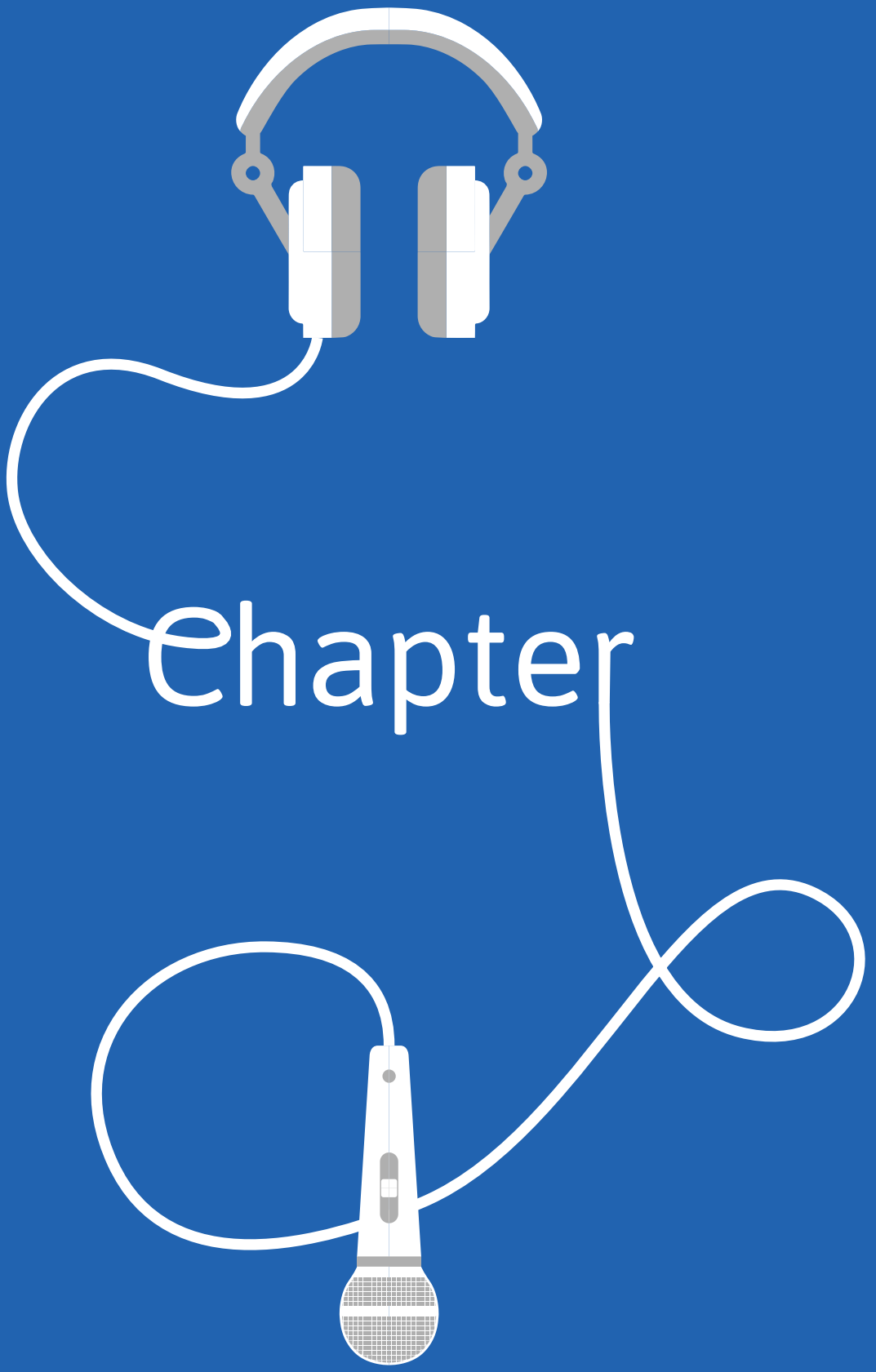
Chapter

APPENDICES

Summary
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Acknowledgements (Dankwoord)
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7

SUMMARY

The ability to read fast and accurate is one of the most fundamental cognitive abilities to be successful at school or to have a successful career. Skilled readers are usually able to identify a written word within 200 milliseconds, and do so without much effort. What often is not realized is that reading is a complex ability that, during development, is built on (speech) motor and linguistic skills. Importantly, humans are not genetically hardwired for reading: reading is a fairly recent cultural invention. Unlike learning to speak, which occurs automatically, learning to read has been called an *'unnatural act',* which requires explicit instruction. Children are taught that the letters of a script (in alphabetic orthographies) correspond with sounds. Particularly during early reading development, reading takes place by overtly producing and concatenating individual letters. Adequate speech skills are therefore often hypothesized to be an essential predictor of reading skills.

A dominant hypothesis in reading research is that the quality of sound representations is related to reading abilities and poor or fuzzy sound representations are frequently associated with dyslexia. The basic idea is that learning to read involves binding letters to sounds and that poorly defined sounds hamper the formation of accurate, fast and stabile letter-sound correspondences. The exact nature and role of the quality of sound representations in reading development and dyslexia is hotly debated. This is partly a consequence of how sound representations are measured in reading research. More specifically, phonological (phonology is the study of sounds) awareness is often used as an indicator of the quality of sound representations and taken as a predictor of reading skills. Phonological awareness is the ability to identify and manipulate individual sounds within words. Questions like 'can you say /spoon/ without the /p/? or can you change the first letters of /Walter Han/? are used to measure phonological awareness. Adequate sound representations are a prerequisite to answer these questions accurately, however, other cognitive abilities, such as attention and working memory are also important. A low performance on phonological awareness tasks, which we consistently observer in individuals with dyslexia, is hence not necessarily an indicator of weak sound representations. Another frequently used method to measure the quality of sound representations is measuring speech perception. In these tasks, participants are usually presented with similarly sounding words, like /pet/ and /bet/, in noisy conditions. The participant should indicate which word was perceived. Speech perception measures are often less strongly correlated with reading skills. Moreover, some researchers claim that speech perception measures require conscious access to sound representations.

Possibly, not the sound representation itself is affected in dyslexia, but individuals with dyslexia could have more difficulties in consciously accessing these representations to accurately answer the question.

Models of the development of speech production suggest that the quality of sound representations is not only reflected in speech perception, but that speech production is also an essential component of sound representations. These models indicate that each sound representations is composed of two different paths. A feedforward path, in which sound representations are mapped onto the motor effectors of the sound, and the feedback path, that monitors whether the produced sound corresponds to the auditory and somatosensory targets of that sound. This feedback path detects abnormalities in the speech production and use them to repair the speech errors. Feedback and/or feedforward paths are updated when the same error is consistently encountered. Monitoring the speech production, as well as using that information to adapt speech, is called the sensorimotor control of speech. This sensorimotor control of speech is particularly important in the early phases of speech development. The route to fluent speaking includes various stages of speech in which, for instance, individual sounds are produced or babbling takes place. Monitoring whether the produced sounds/babbling matches to the intended sound (possibly sounds produced by fluent speakers) is crucial in order to learn speaking. Older children, as well as adults, do not need this sensorimotor control of speech as much as young children do. If you repeatedly produced the same sound in the same way, it becomes unnecessary and inefficient to continuously monitor the production of that sound. Hence, the feedback path of the sensorimotor control of speech might become less active over the years (as a function of the quality of the feedforward path).

Testing the sensorimotor control of speech, and studying how this control relates to reading skills, opens new possibilities to examine the potential role of the quality of sound representations in reading development and dyslexia. Tasks that employ the method of online altered auditory feedback induce a conflict between someone's speech production, and his/her speech perception of that production. The sensorimotor control of speech can be examined with this method. In this summary, I will first explain how an experiment with online altered auditory feedback works, and what it tells about the quality of sound representations. Next, I will describe various study results that were obtained using an altered auditory feedback paradigm. Lastly, I will briefly outline the implications of this research.

Online altered auditory feedback

In my experiments that employ altered auditory feedback a participant should repeatedly produce the same word over and over again (between 95 and 150 times) in a microphone. This microphone is connected with the computer and other hardware, and these systems register the signal, manipulate it, amplify it and feed it back to the participant via the headphone. This all takes place within 10 milliseconds. The amplification of the signal causes the participant to only hear the signal in the microphone and not his/her natural speech. Since the adaptation of the signal happens within 10 milliseconds, and the sound in the headphone sound naturally, participants are assuming to hear his/her own natural speech. In the first phase of the experiment (baseline) the participant produces the same word approximately 25 times, for instance the word /bip/ in the microphone. In this phase, no changes are made to the signal. In the second phase (the ramp phase) the participant produces the word /bip/, multiple times, however, in this phase the signal is gradually changed so that the /i/ in /bip/ starts to sound like an /e/ as in /bep/. In the third phase the adaptation is kept at maximal. Lastly (the after-effect phase), participants produce the /bip/ word for several times again, this time with no adaptations in the signal. Humans usually respond to these manipulations by changing their own speech in the opposite direction of the adaptation. For instance, humans produce a clearer /i/ sound that is farther away from the /e/ sound than usually is the case. This opposing response is particularly strong when the adaptation causes a sound to become another sound.

In these altered auditory feedback experiment, formants in the speech signal are adapted. Formants are resonances at a certain frequency and determine the identity of vowels. The first two formants are particularly important for the identity of the vowel. Een /i/ sound, as in /fit/ has an F1 (i.e. first formatn) of approximately 320 Hz and een F2 of 3200 Hz. Een /e/ sound, as in /pet/ has an F1 of 700 Hz and an F2 of 1800 Hz. If we would increase the first formant of the /i/ vowel in an altered auditory feedback experiment to 700Hz, the resulting sound would be somewhere in between of an /i/ and an /e/. If we additionally decrease the F2 of the /i/ to 1800Hz, the produced /i/ sound would exactly sound like an /e/ vowel. This results in participants saying the word /bip/ in the microphone, while hearing themselves saying the word /bep/.

As mentioned earlier, humans will change their speech in response to this adaptation (again, called the sensorimotor control of speech). This reaction occurs unconscious and without effort. Participants rarely notice changes in the speech signal and a change in speech production could hence be taken as evidence that it's an unconscious, effortless reaction. The strength of the adaptation in response to altered feedback can be indicative of the quality of the underlying sound representation. Imagine, for instance, someone

with a fuzzy definition of how a sound should sound. This person would possibly be very tolerant to alterations in online auditory feedback and, as a consequence, would not strongly react to altered feedback. Another person might lack the precision in the definition of how a sound should be produced. This would of course also result in a more variable speech production, but in relation with the sensorimotor control of speech, this person could well be noticing the alterations in the speech signal, but not be able to make adequate changes to his speech production system. The response to altered auditory feedback can hence reflect the quality of sound representations and is therefore important for dyslexia research. An advantage of using altered feedback is that the response is and unconscious and effortless, and solely reflects the quality of phonological representations. A different response to altered feedback in individuals with dyslexia cannot easily be attributed to, for instance, limited working memory or attention. With respect to the response to altered feedback and dyslexia, I had two competing hypotheses. On the one hand, individuals with dyslexia could respond to a lesser extent to altered feedback than typically reading individuals. If so, a malfunctioning sensorimotor control of speech could be the underlying explanation of their lack of response. On the other hand, individuals with dyslexia could respond to altered feedback stronger than typically reading individuals. This could be caused by very narrow definitions of how a sound should sound (and hence, altered feedback is earlier detected and responded to) or by a flexible motor system for producing sounds (and hence, better able to make changes in the feedforward path. Support for both hypotheses could be found in the literature.

Study results

In Chapter 2, I tested whether dyslexia was characterized by a different response to altered auditory feedback in adults with and without dyslexia. Dyslexia appeared to be related to stronger adaptation under conditions of altered feedback and a weaker de-adaptation when the feedback was back to normal. Moreover, correlational analysis showed that a stronger adaptation and weaker de-adaptation was associated with poorer reading and phonological abilities. I argued that these abnormal responses to altered auditory feedback fit best with a weaker perceptual magnet for sound representations, based on the perceptual magnet theory. This theory claims that a phonetic category prototype functions as an attractor (i.e. magnet) that warps the psychoacoustic space, resulting in poorer discriminability for neighboring stimuli near the category prototype (i.e. a narrower space), and better discriminability farther away from the prototype (i.e. a stretched space). Perturbations in the auditory signal are hence expected to elicit a stronger response when the presented auditory stimulus is farther away from the phonemic category prototype. A weaker magnet would thus

result in a faster deviation from baseline and a slower return to the baseline, exactly the pattern that we observe in individuals with dyslexia. Since the response to altered auditory feedback is unconscious and effortless, I take this as evidence that the sound representations are indeed impaired in individuals with

Chapter 3 examined the neurobiological basis of deficits in the speech network in dyslexia, using three different approaches. First, I measured the structural connectivity between the classical areas of speech perception and production, by means of the arcuate fasciculus. A classical neurobiological model of language associates Wernicke's area with spoken word recognition (Penfield & Roberts, 1959) and Broca's area with speech production (Geschwind, 1965; Levelt, 2013). These areas are structurally connected by the arcuate fasciculus. The results of Chapter 3 show that dyslexia is characterized by a reduced fractional anisotropy of the bilateral arcuate fasciculus, and this likely hampers efficient communication between these regions. Second, I examined whether network quality measures of the functional connectivity in a large and distributed speech network differed between adults with and without dyslexia. Dyslexia appeared to be characterized by a weaker functional integration in this speech network. Lastly, I showed that during speech production, speech sensory areas were hypo-activated in individuals with dyslexia, possibly suggesting a dysfunctional inhibition of self-produced speech monitoring. This set of findings shows that there is a neurobiological basis for a dysfunctional speech organization in dyslexia.

After showing these group differences between adults with and without dyslexia in their behavioral and neural organization of what was hypothesized to reflect the sensorimotor control of speech, in Chapter 4 I attempted to relate individual differences in individuals with dyslexia, with the response to altered auditory feedback and the organization of the arcuate fasciculus. I first replicated the findings described in Chapter 2, by showing that children with dyslexia also responded differently to altered auditory feedback when compared to their typically reading peers. Although the results of Chapter 4 particularly show differences when the altered feedback returns to normal, the same pattern of stronger adaptation under conditions of altered feedback and weaker de-adaptation when the feedback returned to normal was seen in children, as well as it was in adults (see Chapter 2). In addition, individual differences in the severity and persistence of reading difficulties revealed the same patterns, with stronger adaptation and weaker de-adaptation as a function of the severity and persistence of reading difficulties and performance on rapid naming skills, an important precursor of reading. I consider these results as supporting evidence for the claim that dyslexia could be characterized by a weaker sensorimotor magnet. In contrast, a stronger response to altered feedback was associated with better phonological awareness skills in children with dyslexia, and this

was particularly clear in those children who also had a reduced fractional anisotropy in the arcuate fasciculus. This latter finding was attributed to the relatively low importance of phonological awareness skills in reading in a transparent language such as Dutch, while much effort is put in improving phonological awareness skills during treatment.

In Chapter 5, I tested how the sensorimotor control of speech was related to reading development by examining the response to altered auditory feedback in preliterate and literate children and relating this response to reading and reading-related abilities. Moreover, I also investigated whether the transparency of the orthography has an impact on the presence and strength of these relations, by including American and Dutch children. In Dutch, the mapping between letters and sounds is fairly consistent and Dutch is therefore a transparent orthography. For example, the letter combination 'ee' (as in 'been', 'scheen') is almost always pronounced in the same way (although exceptions exist). In contrast, the mapping between letters and sounds is very inconsistent in English. For example, the letter combination 'ea' (as in 'head', 'heat', 'heart', 'year', 'break', 'earth') all show different pronunciations of the 'ea' combination. Since the relation between letters and sounds is less consistent in English, I expected a weaker relation between the sensorimotor control of speech in English. A total of 225 (Dutch 148: American 77) children from 4-8 years old participated in an altered auditory feedback experiment and completed tests measuring reading and reading-related skills. I found that the response to altered auditory feedback was related to being able to read, with a stronger response to altered auditory feedback for the literate children. Additionally, precursors of reading ability in the preliterate children were also positively related with the strength of the response to altered feedback. I related these findings to the development of the vowel space in young children. In adults, it has been shown that the strength of the response to altered feedback depends on whether the alteration changes the vowel close to the center or to the border of the vowel category. With increasing age, the vowel definitions are becoming more compact and separate from other vowel categories. Hence, the applied vowel alterations were probably closer to the border of the vowel category, resulting in stronger adaptation for the literate children. These changes in the vowel space co-occur with, and possibly facilitate, the integration of orthography into phonetic representations.

Conclusion

In this dissertation, I provided evidence that the sensorimotor control of speech is associated with reading skill. Children and adults with dyslexia showed a stronger adaptation response to altered auditory feedback and a weaker de-adaptation response when the feedback was back to normal. Neuroimaging measures further

supported that dyslexia could be related to a deficient sensorimotor control of speech. The results of the presented thesis showed that dyslexia is characterized by impaired phonological representations, and describe the possible nature of this impairment: a weaker perceptual magnet for sound representations. The altered auditory feedback paradigm allows to directly compare contradicting theories of dyslexia and provides an interesting and challenging new window for future dyslexia research.

Considering the importance of literacy skills for an individual's academic and economic prospects as well as the societal costs associated with low literacy skills, there is probably no need to convince anyone of the relevance of research into the origins of differences in reading skill. It should be noted that, considering the many questions that this thesis is generating, the results of this thesis are not directly applicable to improve reading skills of clinical or general populations. However, I am convinced that the main interpretation (weaker magnet in dyslexia) and/or the used methodologies (measuring sensorimotor control) will help the field move forward which could ultimately result in advancements in dyslexia prevention, assessment and intervention.

NEDERLANDSE SAMENVATTING

Het vermogen om snel en accuraat te kunnen lezen is één van de belangrijkste cognitieve vaardigheden om succesvol te kunnen zijn op school of om een succesvolle carrière te hebben. Bedreven lezers zijn gewoonlijk in staat om, zonder moeite, binnen 200 milliseconden een geschreven woord te herkennen. Het gemak waarmee lezen gepaard gaat, in bedreven lezers, is extra bijzonder als we bedenken dat lezen een redelijk recente culturele uitvinding is die gebaseerd is op spraak en taalvaardigheden. Mensen zijn niet genetisch 'gebouwd' om te kunnen lezen: in tegenstelling tot bijvoorbeeld leren praten, wat automatisch gebeurd bij voldoende blootstelling aan spraak, is leren lezen bijna alleen mogelijk door middel van expliciete instructie. Aan kinderen wordt geleerd dat de letters van het schrift (in alfabetische schriften zoals het Nederlands) klanken representeren. Zeker in de beginfase van het leren lezen worden letters ook hardop uitgesproken en aan elkaar geplakt om een woord te vormen. Goede spraakvaardigheden worden daarom essentieel geacht om goed te kunnen leren lezen.

Een dominante hypothese in het leesonderzoek is dat de kwaliteit van klankrepresentaties gerelateerd is aan leesvaardigheden en slechte, onduidelijk gedefinieerde klankrepresentaties worden herhaaldelijk geassocieerd met dyslexie. Het idee erachter is dat leren lezen een proces is waarin letters aan klanken gekoppeld moeten worden, en dat slecht gedefinieerde klanken een goede, snelle en stabiele koppeling tussen letters en klanken in de weg staat. Over de precieze aard en rol van klankrepresentaties in leesontwikkeling en in dyslexie wordt echter hevig gedebatteerd en dit heeft onder andere te maken met hoe klankrepresentaties gemeten worden. In de literatuur over voorspellers van leesvaardigheden wordt fonologisch (fonologie is de studie van spraakklanken) bewustzijn vaak gebruikt als indicator van de kwaliteit van klankrepresentaties. Fonologisch bewustzijn is het vermogen om klanken binnen gesproken woorden te kunnen onderscheiden en te manipuleren en wordt bijvoorbeeld gemeten met vragen als: 'Kun je het woord /klap/ zeggen zonder de /l/?' of 'Kun je de eerste letters van /Kabouter Plop/ omdraaien?' (antwoord: /Pabouter Klop/). Om dit soort vragen te kunnen beantwoorden zijn goede klankrepresentaties wel nodig, maar andere cognitieve vaardigheden, zoals aandacht en werkgeheugen, zijn ook van belang. Beneden gemiddelde scores op fonologisch bewustzijn, die we consequent zien bij kinderen met dyslexie, betekenen dan ook niet noodzakelijkerwijs dat de klankrepresentaties niet adequaat zijn. Een andere veelgebruikte methode om de kwaliteit van klankrepresentaties is het meten van spraakwaarneming. In dit soort taken worden bijvoorbeeld woorden die erg op elkaar lijken, zoals /bak/ en /pak/, in slecht hoorbare omstandigheden afgespeeld, waarbij een participant moet aangeven welk woord gehoord is. Dit soort taken laat echter vaak een minder sterke relatie zien

met leesvaardigheden en/of dyslexie. Daarnaast zijn er onderzoekers die ook dit geen adequate meting vinden van de kwaliteit van klankrepresentaties, onder andere omdat er een bewuste 'toegang' tot klankrepresentaties nodig is om goed te kunnen presteren op dit soort taken. Mogelijk is niet de kwaliteit van de representatie zelf aangetast in dyslexie, maar hebben mensen met dyslexie meer moeite met het bewust benaderen van deze representaties om vervolgens een keuze te kunnen maken.

Modellen van hoe spraakproductie zich ontwikkelt geven echter aan dat de kwaliteit van klankrepresentaties niet alleen gereflecteerd wordt in de waarneming van spraak, maar dat ook de productie van spraak een cruciaal onderdeel is van een goede klankrepresentatie. Deze modellen geven aan dat elke klankrepresentatie is opgebouwd uit twee paden. Een 'feedforward' pad, waarin de klankrepresentatie aan de productie van een klank gekoppeld wordt, en een 'feedback' pad, die detecteert of de geproduceerde klank overeenkomt met de auditieve en somatosensorische doelen van die klank. Dit feedback pad detecteert afwijkingen en dit wordt gebruikt om spraakfouten te herstellen. Wanneer dezelfde fout consistent voorkomt, wordt het feedforward en/of feedback pad bijgewerkt. Dit monitoren van de spraakproductie, en het gebruiken van deze informatie om spraak productie aan te passen, wordt de sensomotorische controle van spraak genoemd. Deze controle is vooral belangrijk in de eerste jaren van de ontwikkeling. In die eerste jaren doorlopen kinderen verschillende stadia, bijvoorbeeld het uitstoten van losse klanken en brabbelen, om uiteindelijk goed te kunnen praten. Oudere kinderen en volwassenen hebben deze sensomotorische controle van spraak veel minder nodig. Als je oneindig vaak dezelfde klank op dezelfde manier hebt geproduceerd en het telkens goed ging, is het minder nodig om voortdurend en gedetailleerd te controleren of de uitspraak wel overeenkwam met de gestelde doelen. Het feedback pad van de sensomotorische controle van spraak wordt dan ook mogelijk minder actief in de loop der jaren.

Het testen van de werking van de sensomotorische controle van spraak, en het onderzoeken van hoe deze controle relateert aan leesvaardigheden, geeft nieuwe mogelijkheden om te onderzoeken of, en hoe, klankrepresentaties gekoppeld zijn aan leesontwikkeling en dyslexie. Taken waarin de methode van online aangepaste auditieve feedback worden gebruikt, induceren een conflict tussen wat iemand produceert en wat diegene zichzelf hoort zeggen. Op deze manier kan de werking van de sensomotorische controle getoetst worden. In het vervolg van deze samenvatting werk ik eerst uit hoe een experiment met aangepaste auditieve feedback werkt, en wat het zegt over de kwaliteit van klankrepresentaties. Daarna bespreek ik de verschillende

onderzoeksresultaten die, mede met behulp van experimenten met aangepaste auditieve feedback, gevonden zijn. Als laatste werk ik kort uit wat de implicaties zijn van dit onderzoek.

Online aangepaste auditieve feedback

In mijn experimenten waarin gebruik wordt gemaakt van online aangepaste auditieve feedback, spreekt een proefpersoon hetzelfde woord meerdere malen (tussen de 95 en 150) door een microfoon. Deze microfoon is aangesloten op een computer en andere hardware die het signaal registreren, eventueel bewerken, versterken, en binnen 10 milliseconden laten horen door de koptelefoon. Door de versterking van het geluid hoort een proefpersoon niet zichzelf praten, maar hoort hij alleen wat in de koptelefoon afgespeeld wordt. Omdat de aanpassing in zo'n korte tijd gebeurd (<10 ms) en het geluid door de koptelefoon heel natuurlijk klinkt, is elke participant in de veronderstelling dat hij/zij gewoon zichzelf hoort praten. In de eerste fase van elk experiment (de baseline fase) spreekt de participant ongeveer 25 keer hetzelfde woord, bijvoorbeeld /bip/, door de microfoon en wordt dat signaal niet aangepast en gewoon afgespeeld in de hoofdtelefoon. In de tweede fase (de ramp fase) spreekt de participant opnieuw meerdere malen het woord /bip/ uit in de microfoon. Echter, bij elke volgende trial wordt het signaal iets aangepast, zodat de /i/ in /bip/ steeds meer als een /e/ begint te klinken. In de derde fase (de hold fase) wordt de aanpassing constant op een hoog niveau gehouden. Daarna (in de ramp down of after-effect fase) wordt de manipulatie weer afgebouwd en spreekt de participant nog een aantal keer het woord /bip/ in de microfoon zonder aanpassing. Mensen zijn goed in staat om op dit soort aanpassingen te reageren en de eigen spraak aan te passen zodat de manipulatie in het signaal weer (deels) ongedaan wordt gemaakt. Mensen gaan bijvoorbeeld nog duidelijker een /i/ klank produceren die minder lijkt op de /e/ klank dan normaal al het geval is. Mensen reageren vooral sterk op de aanpassing als de manipulatie ervoor zorgt dat de klank erg gaat lijken op een andere klank.

In al dit soort experimenten waarin gebruik wordt gemaakt van aangepaste feedback, worden formanten in het spraaksignaal aangepast. Formanten zijn resonanties op bepaalde frequenties die de identiteit van een klinker bepalen. Met name de eerste twee formanten zijn cruciaal voor de identiteit van de klinker. Een /i/ klank, zoals in /kip/, heeft een F1 (i.e. eerste formant) van ongeveer 320 Hz en een F2 van 3200 Hz. Een /e/ klank, zoals in /bed/, heeft een F1 van 700 Hz en een F2 van 1800 Hz. Als we in een experiment de eerste formant van de /i/ klank zouden verhogen naar de eerste formant van de /e/ klank krijg je een klank die ergens tussen de /i/ en de /e/ in zit. Als we dan ook de tweede formant van de /i/ gaan verlagen naar de tweede formant van de /e/ dan

krijg je een klank die precies klinkt als een /e/. Dat zorgt ervoor dat participanten dus het woord /bip/ kunnen uitspreken in de microfoon, maar zichzelf echt het woord /bep/ horen zeggen.

Zoals gezegd gaan mensen corrigeren als datgene wat ze horen anders is dan wat ze bedoelen uit te spreken (nogmaals, een mechanisme dat de sensomotorische controle van spraak heet). Deze reactie gebeurt onbewust en zonder enige moeite. Mensen hebben zelden in de gaten dat het spraaksignaal aan wordt gepast en passen hun eigen spraak dus ook zonder moeite aan. De sterkte van de aanpassing kan veel zeggen over de kwaliteit van klankrepresentaties. Stel bijvoorbeeld dat iemand niet zo een scherpe definitie heeft van hoe een bepaalde klank moet klinken. Zo iemand is redelijk 'tolerant' voor verschillende uitspraken van een /i/ klank en zou dus redelijk laat gaan reageren op aangepaste feedback. Iemand kan ook niet zo een scherpe definitie hebben van hoe een klank uitgesproken moet worden. Dat is allereerst natuurlijk te zien in hoeveel variatie er zit in iemands uitspraken in de baseline fase. Daarnaast, in termen van de sensomotorische controle van spraak, zou zo iemand misschien wel horen (nogmaals, onbewust) dat de spraak aangepast is, maar niet voldoende in staat zijn om adequate motorische aanpassingen te doen aan hun spraaksignaal. De reactie op aangepaste feedback kan dus veel zeggen over de kwaliteit van klankrepresentaties en is daarom van belang voor onderzoek naar dyslexie. Een bijkomend belangrijk voordeel van het gebruik van aangepaste feedback is dat de reactie onbewust en zonder moeite gaat. Een afwijkende reactie op aangepaste feedback van mensen met dyslexie, ten opzichte van goede lezers, kan dus niet/moelijker geweten worden aan een gebrek in bijvoorbeeld het werkgeheugen. Voorafgaand aan de verschillende onderzoeken waren er ruwweg twee logische mogelijkheden voor de reactie op aangepaste feedback van mensen met dyslexie. Enerzijds zouden mensen met dyslexie minder sterk kunnen reageren op aangepaste feedback. Dat zou kunnen komen door een minder goede werking van de sensomotorische controle van spraak. Dat zou kunnen resulteren in minder scherpe definities van hoe een klank moet klinken of minder goede motorische commando's voor klanken. Anderzijds zouden mensen met dyslexie juist te sterk kunnen reageren op aangepaste feedback. Dat zou veroorzaakt kunnen worden door een overgevoelige definitie van hoe een klank zou moeten klinken, of 'flexibele' motorische commando's voor klanken. Beide mogelijkheden hebben goede onderbouwingen van eerder gedaan onderzoek.

Bevindingen onderzoek

In hoofdstuk 2 heb ik, bij volwassenen met en zonder dyslexie, onderzocht of dyslexie gekenmerkt wordt door een andere reactie op aangepaste auditieve feedback. Volwassenen met dyslexie lieten een sterkere aanpassing van de spraak zien tijdens de aangepaste auditieve feedback en een zwakkere terugkeer naar hun eigen 'baseline' wanneer de auditieve feedback weer normaal werd. Daarnaast lieten de analyses ook zien dat een sterkere aanpassing en een zwakkere terugkeer samenhingen met zwakkere lees- en fonologische vaardigheden. Ik heb beargumenteerd dat deze abnormale reactie op aangepaste auditieve feedback het best verklaard kan worden door een zwakkere perceptuele 'magneet' voor klankrepresentaties, gebaseerd op de *perceptual magnet* theorie. Deze theorie geeft aan dat het prototype van een klankrepresentatie fungeert als een soort magneet die de waarnemingsruimte vervormd. Dit zorgt voor minder discriminatievermogen voor stimuli die dicht bij het prototype ligt en een beter discriminatievermogen voor stimuli die daar verder vandaan liggen. Aanpassingen in auditieve feedback hebben, in de lijn van deze theorie, dus een grotere impact naarmate de aanpassing ervoor zorgt dat de stimulus verder bij het prototype vandaan komt. Een zwakkere 'magneet' zorgt er dus voor een snellere afwijking van de baseline en een langzamere terugkeer naar de baseline, precies het patroon dat we bij mensen met dyslexie zien. Omdat de reactie op aangepaste auditieve feedback volledig onbewust en zonder moeite gebeurd zie ik dit als bewijs dat de klankrepresentaties bij mensen met dyslexie inderdaad minder goed zijn dan bij mensen zonder dyslexie.

In hoofdstuk 3 heb ik met behulp van drie verschillende benaderingen, bekeken of er een neurobiologische basis is voor tekorten in het spraaknetwerk bij mensen met dyslexie. Eerst heb ik gemeten of de structurele verbindingen die gebieden voor spraakwaarneming en spraakproductie met elkaar verbinden, zwakker zijn bij mensen met dyslexie. Deze structurele verbinding heet de arcuate fasciculus. De resultaten van mijn onderzoek geven aan dat deze arcuate fasciculus bij mensen met dyslexie in beide hersenhelften zwakker zijn dan bij gemiddelde lezers. Als tweede heb ik bekeken of, en hoe, de functionele verbindingen tussen hersengebieden die betrokken zijn bij spraak gerelateerd zijn aan dyslexie. Functionele connectiviteit kan gemeten worden door te bekijken hoe activiteit in één hersendeel de activiteit in een ander deel voorspelt. Dyslexie blijkt te worden gekenmerkt door minder goede functionele verbindingen tussen gebieden die met spraakwaarneming en productie te maken hebben. Als laatste heb ik bekeken welke hersengebieden meer of minder geactiveerd worden tijdens het waarnemen en produceren van spraak. Tijdens het produceren van spraak activeren mensen met dyslexie hun spraakwaarneming gebieden in mindere mate dan

gemiddelde lezers. Als geheel denk ik dat deze combinatie van bevindingen laat zien dat er een neurobiologische basis is voor een tekort in de organisatie voor spraak bij mensen met dyslexie.

Nadat ik deze groepsverschillen tussen mensen met en zonder dyslexie in de sensomotorische controle van spraak heb laten zien, heb ik in hoofdstuk 4 onderzocht of individuele verschillen in kinderen met dyslexie gerelateerd zijn aan deze sensomotorische controle van spraak en wat de rol van de arcuate fasciculus daarbij is. Individuele verschillen in de ernst en mate van behandelbaarheid van dyslexie relateerden aan de sterke van de reactie op aangepaste auditieve feedback. Met andere woorden, kinderen die meer moeite hadden met lezen, en minder reageerden op de aangeboden dyslexiebehandeling, veranderen hun spraak meer tijdens aangepaste auditieve feedback en kwamen minder terug op hun oorspronkelijke baseline wanneer de aanpassing in de feedback weer ongedaan was gemaakt. Opmerkelijk genoeg relateerde een sterkere reactie op aangepaste feedback ook positief met fonologische vaardigheden, en dit was vooral het geval bij die kinderen met een zwakke arcuate fasciculus. We vermoeden dat deze laatste bevinding te maken heeft met het relatief lage belang van fonologisch bewustzijn in het lezen van Nederlands (in tegenstelling tot bijv. Engels) terwijl er wel, gedurende de behandeling, veel energie wordt gestoken in het verbeteren van deze fonologische vaardigheden.

In het laatste onderzoek van dit proefschrift, beschreven in hoofdstuk 5, heb ik onderzocht in hoeverre de sensomotorische controle van spraak samenhangt met leesontwikkeling van kinderen die nog niet kunnen lezen en die net kunnen lezen. Daarnaast heb ik ook onderzocht of de transparantie van het schrift de relatie tussen sensomotorische controle van spraak en leesontwikkeling verandert. De transparantie van het schrift geeft aan hoe consistent letters dezelfde klanken representeren. In het Nederlands wordt letters heel vaak op dezelfde manier uitgesproken en het Nederlands is daarmee een redelijk transparant schrift. Bijv. de lettercombinatie 'ee' (als in 'been', 'scheen' etc.) wordt praktisch altijd op dezelfde manier uitgesproken. Het Engels heeft veel meer variatie in hoe letters uitgesproken worden en is daarmee een niet transparant schrift. De lettercombinatie 'ea' (als in 'head', 'heat', 'heart', 'year', 'break', 'earth') kan op veel verschillende manieren worden uitgesproken. De relatie tussen schrift en spraak is dus in het Engels minder groot en ik verwachtte dan ook een minder sterke relatie tussen de sensomotorische controle van spraak en leesontwikkeling in het Engels dan in het Nederlands. 225 (148 Nederlandse en 77 Amerikaanse) kinderen van 4-8 jaar hebben deelgenomen aan dit onderzoek waarin de reactie op aangepaste auditieve feedback en verschillende (voorspellers van) leesvaardigheden werden gemeten. Deze studie liet zien dat kinderen die konden lezen een sterkere reactie lieten zien op aangepaste

auditieve feedback dan kinderen die nog niet konden lezen. In overeenstemming met de verwachting was dit sterker het geval bij de Nederlandse kinderen dan bij de Amerikaanse kinderen. Binnen de groep kinderen die nog niet konden lezen werden er ook sterkere reacties op aangepaste feedback waargenomen bij die kinderen met hogere scores op voorspellers van leesvaardigheden. Deze bevindingen hebben waarschijnlijk te maken met de ontwikkeling van de klankdefinities in kinderen. Naarmate kinderen ouder worden, worden de klanken steeds scherper en preciezer gedefinieerd. Dit kan zorgen voor een sterkere reactie op aangepaste auditieve feedback. Deze veranderingen in de klankdefinities gebeuren gelijktijdig, en faciliteren mogelijk, de leesontwikkeling.

Conclusie

De hoofdconclusie van dit proefschrift is dat de sensomotorische controle van spraak en leesvaardigheden aan elkaar gerelateerd zijn. Kinderen en volwassenen met dyslexie passen hun eigen spraak (te) snel aan bij aangepaste auditieve feedback en komen (te) langzaam terug bij hun eigen baseline wanneer de feedback weer normaal is. De hersenbeeldvormingsmaten ondersteunden de bevinding dat dyslexie gerelateerd is aan een beperkte sensomotorische controle van spraak. De resultaten van dit proefschrift laten zien dat dyslexie gekarakteriseerd wordt door een beperkte kwaliteit van klankrepresentaties, en beschrijven de mogelijke aard van de beperking: een zwakkere perceptuele magneet voor klankrepresentaties. Het gebruik van aangepaste auditieve feedback helpt om verschillende theorieën over dyslexie verder te onderzoeken en geeft veel nieuwe mogelijkheden voor onderzoek naar dyslexie.

Gezien het belang van leesvaardigheden voor iemands academische en economische vooruitzichten, alsmede de maatschappelijke kosten die geassocieerd zijn met laaggeletterdheid, hoef ik waarschijnlijk niemand te overtuigen van het nut van onderzoek naar de oorzaak van verschillen in leesvaardigheden. De resultaten van dit proefschrift zijn, gezien de vele vragen die de resultaten oproepen, niet direct toepasbaar om leesvaardigheden te verbeteren. Echter, de belangrijkste interpretatie (een zwakkere magneet in dyslexie) en/of de gebruikte methoden (het meten van de sensomotorische controle van spraak door middel van aangepaste auditieve feedback) zullen het onderzoek naar dyslexie verder helpen en kan uiteindelijk resulteren in vooruitgang in de preventie, diagnostiek en interventie van dyslexie.

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CURRICULUM VITAE

Mark van den Bunt (Amerfoort, 27 maart 1989) holds a bachelor degree in Psychology and master degrees in Neuropsychology and in Neuroscience & Cognition (Research master). During his studies, Mark became intrigued by the complex relation between brain and behavior, and fascinated by the consequences of impaired brain functioning. After various internships in clinical neuropsychology Mark decided a solid base in research would be the best basis for a career in- or outside neuroscience. In 2013, Mark began a PhD project to unravel the relation between speech and reading skills. This project was part of an international collaboration (Haskins Laboratories, USA; Normal Beijing University, China; University of Warsaw, Poland & Radboud University, The Netherlands), aimed to examine whether predictors of reading development are universal or language-specific. Mark taught in the educational sciences programmed and supervised several bachelor and master student's in conducting research and writing their thesis. He visited two laboratories (Basque Center for Brain, Cognition and Language, Spain; Haskins Laboratories, Yale University, USA) to discuss analyses and present results and he presented his findings at multiple national and international conferences. At the time of writing, Mark is working as a Research Consultant at Zilveren Kruis, a health insurance company.

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